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DUMB-BELL GANGLIONEUROMATA OF THE SPINE WITH A REPORT OF FOUR CASES

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DUMB-BELL tumours of the spine are relatively uncommon. Eden (1941) reviewed 234 spinal tumours seen at the National Hospital, Queen Square, and at University College Hospital in the previous 20 years and found 32 dumb-bell tumours amongst them. Amongst these there was included only one dumb-bell ganglioneuroma.

Heuer (1929) in a classic survey of the literature on dumb-bell tumours collected 61 cases and added 3 more. Amongst these 64 cases there were only 5 ganglioneuromas. Naffziger and Brown (1933) reported 15 dumb-bell tumours, but only one was a ganglioneuroma.

It would appear, therefore, that dumb-bell ganglioneuroma is a very rare tumour, for, considering Heuer's and Eden's cases together, among a total of 96 dumb-bell tumours of the spine, only 6 were ganglioneuroma (an incidence of just over 6 per cent). In reviewing the available literature we have been able to discover only 11 cases, including those mentioned above.

Ganglioneuroma of the non-dumb-bell type is, of course, a much commoner lesion. Thus Stout (1947) recorded that 250 cases were described and added 10 cases of his own, a total of 260. We can find reports of only 9 cases of the dumb-bell variety of ganglioneuroma up to that year, a relative incidence of only 3·5 per cent as compared with non-dumb-bell ganglioneuroma.

Dumb-bell ganglioneuromata seem to be relatively more frequent in the dorsal region. Thus Rogers and Keogh (1950) brought the total of reported cases of intrathoracic ganglioneuroma to 90. Of these, 8 were dumb-bell in form, giving a relative incidence of 9 per cent in the dorsal region. Three years later this had apparently increased to 11 per cent by the addition of a further case of mediastinal ganglioneuroma with dumb-bell features (House and Bahrend, 1953).

In this paper we present 4 further cases of ganglioneuroma all of which were producing cord compression and all of which were 'dumb-bell' in accordance with the description on p. 314.

Our own case reports will be preceded by a description of the 11 previously recorded cases of dumb-bell ganglioneuroma.

REVIEW OF THE LITERATURE

1. Loretz (1870).—Made the first report of a patient with a dumb-bell spinal tumour appearing to be a ganglioneuroma.

This concerned a woman of 35 who died shortly after admission to hospital in an epileptic fit. The post-mortem examination revealed a tumour the size of a hen's egg in the superior mediastinum at the level of the second and third ribs. This had an intraspinal extension. The histological examination showed a large number of ganglion cells growing in clumps and also singly, with many non-myelinated and a few myelinated nerve-fibres. Loretz believed this tumour to be a ganglioneuroma developed from hyperplastic enlargement of a normal ganglion.

2. Busse (1898).—Recorded the case of a boy of 4 with this condition. This represents the first description of a ganglioneuroma discovered during life. The patient had had paraparesis since infancy; he was also incontinent of urine and fæces. On examination a tumour mass was palpable in the left hypochondrium. The diagnosis was glioma or fibroma in the dorsal and upper lumbar regions with intraspinal extensions involving the lumbar portion of the spinal cord. The paravertebral portion was removed and found to weigh 700 g. A portion about the size of an egg was left due to collapse of the patient during operation. Microscopic examination revealed a ganglioneuroma which was thought to have originated from a lumbar sympathetic ganglion.

The wound healed by first intention and some time later a decrease in the degree of paralysis of the legs was noted. No separate attempt was made to remove the intraspinal portion. Subsequent progress was not recorded.

3. Rapp (1913).—Described a similar case to the foregoing. This was a boy of 4 who, since the age of one year, had developed complete paraplegia with total sensory loss resulting in marked decubitus ulcers. Urinary and fæcal incontinence was present and also paralysis of the lower back and abdominal muscles. In the lumbar region, to the right of the spine, was a soft tumour. The main part of this was removed, the intraspinal component being removed at the same time with division and suture of the dura. Histological examination revealed features mainly of a ganglioneuroma, but there was in addition infiltration of muscles. Rapp stated that it could justifiably be called a mixed tumour, concluding nevertheless that it was a true ganglioneuroma. There was some improvement in

sensation following the operation, but no real recovery of motor power. No further follow-up is recorded.

4. Stout (1924).—Reported a patient, a boy of 2½, who at the age of 7 months developed abdominal pain and unsteadiness of gait with a right foot-drop. When he was 2 years old, the weakness increased, the child was unable to sit up, and sphincter control was lost. The chest radiograph revealed a dense shadow in the upper right mediastinum. A needle biopsy was performed and ganglion cells demonstrated in the specimen. Seven months later a laminectomy was done by Elsberg and Neuhof and a long, encapsulated extradural tumour was removed from the upper dorsal and lower cervical regions. This had the histological appearance of a ganglioneuroma. This is the first report of a laminectomy being performed for a tumour of this nature.

5. Cushing and Wolbach (1927).—Reported the case of a boy of 12 who at the age of 2 years developed a paravertebral swelling at the mid-thoracic level. A clinical diagnosis of sarcoma was made, but on exploration the tumour proved to be a cellular sympathicoblastoma (neuroblastoma). This had arisen from the region of an intervertebral foramen and had extended into the spinal canal and the spinal muscles.

Coley's toxins were given and the activity of the tumour subsided. However, a paraplegia appeared which persisted in spite of some improvement over a period of a few years. At laminectomy a few years later, there was revealed a relic of the former growth. The cells of this had become completely differentiated into ganglion, capsular, and neurilemma cells. This, in Cushing's opinion, illustrated that a sympathetic neuroblastoma may be a precursor of a ganglioneuroma.

6. Naffziger and Brown (1933).—In their paper on 15 hour-glass tumours they report the case of a boy of 7 who presented with paraplegia. This developed rapidly over two days and there was a previous history of occasional pain in the chest for three years. On examination he had a bulge below the right scapula and a complete flaccid paraplegia with sensory loss up to the 8th thoracic segment. Radiographs showed destruction of the transverse process of the 6th thoracic vertebra on the right side and a large paravertebral swelling from the 5th to the 10th ribs. A correct clinical diagnosis of hour-glass tumour was made although the radiographic diagnosis was that of an inflammatory process.

At laminectomy, a tumour was found extending into the muscles of the back and around the vertebral bodies into the chest. Its intraspinal portion had a more vascular appearance and formed an extradural tumour extending from the 3rd to the 7th thoracic vertebræ. The histological diagnosis on this occasion was hæmangio-endothelioma.

Recurrence of the tumour led to further laminectomy two months later, in spite of which the child deteriorated during the ensuing three months and died. Autopsy revealed a recurrence of the growth communicating with an enormous thoracic tumour filling almost the entire right side of the chest cavity. This was firm, nodular, and encapsulated. The final pathological diagnosis was ganglioneuroma of sympathetic origin.

7. Eden (1941).—In his paper on dumb-bell tumours of the spine he described the case of a man of 23 with weakness of the legs and difficulty in micturition of three weeks' duration. When 5 years old he had been admitted to another hospital with a similar complaint and a radiograph at that time revealed an opacity in the chest which was diagnosed as a sarcoma of the spine and judged to be inoperable. He had recovered from his paralysis in a few years and had led a normal life until the onset of paralysis for the second time. On examination there were signs of a left-sided intrathoracic tumour and a severe spastic paraplegia with a sensory level at the 5th thoracic segment. A radiograph showed a large semilunar tumour in the left upper mediastinum and a growth in the foramen between the 4th and 5th thoracic vertebræ. Thoracotomy was performed and a large lobulated tumour arising from the spine by several stalks was encountered. Before removal could be completed, the operation had to be abandoned and the patient died shortly afterwards. At autopsy there were several lobes of tumour extending along the spinal canal and these had caused severe compression of the cord. The intraspinal portion of the tumour was intimately related to several nerve roots. Histological examination of the tumour revealed a dense network of nerve-fibres with here and there groups of ganglion cells.

8. Paterson and Pilcher (1941).—Described the case of a boy of 4 who had suffered from asthma since the age of one year. There were no neurological symptoms. Radiography revealed a rounded opacity on the right side of the posterior mediastinum with widening of the 4th intercostal space and erosion of the 4th rib. There were no abnormal signs in the central nervous system. At thoracotomy the tumour was removed and a small prolongation found to enter the spinal canal through the 4th intervertebral foramen. The pathological examinations were carried out by Professor Nevin; the histological appearances were those of a ganglioneuroma and a photomicrograph was included in the report.

9. Sames (1950).—Recorded an example occurring in a girl of 3 years. She presented with paraplegia developing over two months. This was almost complete and extended to the 4th thoracic segment. Radiographs showed a large, partially calcified tumour in the right posterior mediastinum. A diagnosis of dumb-bell tumour was made and a laminectomy performed. This revealed an extensive growth of a large, lobulated, extrathecal tumour situated mainly in front of the cord and producing a gross compression of it. The tumour was connected with the main paravertebral mass via a right intervertebral foramen. There were also small prolongations into the 3rd and 4th foramina on the right and into the 4th and 5th on the left. On microscopic examination, well-developed ganglion cells containing neurofibrils were seen in a dense network of axis cylinders. Myelin sheaths were not demonstrated.

A 5-year follow-up examination was obtained in this case. There was residual spasticity of the legs but no evidence of recurrence.

10. Rogers and Keogh (1950).—Described the case of a woman of 26 presenting with left arm and shoulder pain for six weeks. Radiology revealed a large mass in the upper mediastinum and a clinical diagnosis of neurofibroma or ganglioneuroma was made. There were no neurological signs.

On thoracotomy the tumour was found to arise from the first four thoracic sympathetic ganglia and had a prolonged extension into the 2nd intervertebral foramen. This is the only reference to its dumb-bell character. The histological appearances of the tumour substantiated the diagnosis of ganglioneuroma.

11. House and Bahrend (1953).—Reported the case of a man of 64 presenting with a history of cough and paroxysmal dyspnœa for one year. On radiographic examination there was found a left-sided tumour behind the heart associated with enlargement of the intervertebral foramina. A myelogram revealed an extradural defect on the left side at the level of the 10th thoracic vertebra. The thoracic tumour was removed through the bed of the 10th rib together with a small prolongation which entered the extradural space through the intervertebral foramen. On histological examination these were bundles of nerve-fibres with interspersed ganglion cells. The patient made a good recovery.

CASE REPORTS

Case 1.—G. R. S. (M.V.H. 30284). Male, aged 53.

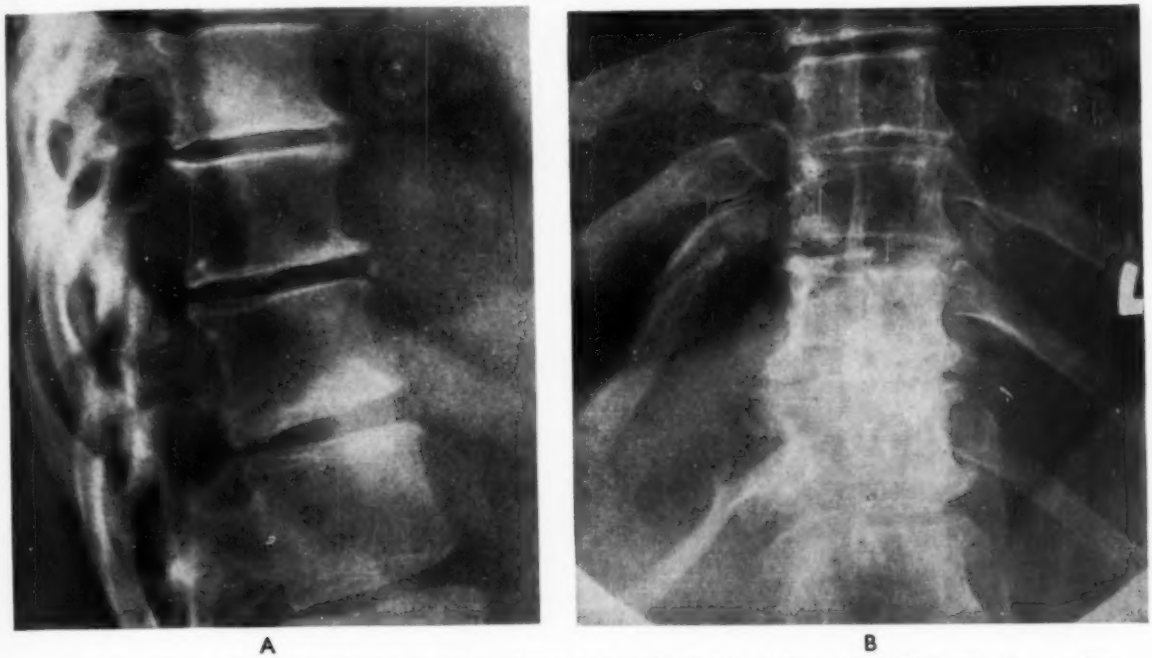
This patient, a clergyman, was admitted to Maida Vale Hospital under the care of Dr. Nevin in November, 1953, with the following complaints: (1) Weakness of his legs for 34 years; (2) Pains in the back for many years; (3) Bladder symptoms for 4 years.

HISTORY.—*Motor Weakness.*—Mild symptoms were first noticed in 1917. They consisted of a sensation of weakness in the back, stiffness and weakness of the legs. Four years later a diagnosis of disseminated sclerosis was made, but there was little change in his condition for 22 years. Then in 1939 a remarkable improvement occurred allowing him to dispense with his walking-stick and to take up gardening. However, his legs did not fully recover and in 1944 he was examined by a neurologist and a full investigation was carried out. A diagnosis of disseminated sclerosis was again made though with some reluctance. His liver was found to be enlarged; this was said to be due to a hepatoma. From 1947 his condition deteriorated, the weakness and spasticity of his legs increasing, being always more marked on the left. Between this year and the middle of 1953 he had two falls, each of which led to confinement to bed for 2–3 months with further deterioration. At the time of his admission to hospital he could walk a distance of 100 yards only and required the aid of two sticks.

Pain.—He had experienced two types of pain. The first was a dull ache felt diffusely in the back, associated with a feeling of weakness, but without radiation. It had occurred intermittently since 1919. The second was a burning pain felt in the midline of the small of the back, coming on for short periods of about ten minutes, and without radiation or aggravation by coughing. It was first felt in 1923 and had occurred very occasionally since.

Sphincter Disturbance.—Symptoms of disturbance of bowel and bladder control were slight but progressive, since they were first noted in 1949. Impotence had been present during the last five years.

Sensory Involvement.—The symptoms of impairment of sensation were overshadowed by the motor manifestation, and to some extent direct inquiry was required to elicit them. They consisted of disturbance of proprioception, there being an unawareness of the position of his feet, and where his knees were on the hassock while kneeling, etc. In addition he had noticed a sensation of mild tingling in the feet. Further inquiry revealed that he had never been really active. As a child he was never able to run or jump well. There was no history of root pains and no symptoms to suggest retrobulbar neuritis, involvement of the upper limbs, or of cranial nerve, special sense, or cerebral involvement. The family history and previous medical history were not contributory to the present inquiry.



A B

FIG. 347.—*Case* 1. Lower dorsal spine showing erosion of the posterior surface of the bodies of D.10 and D.11. Enlargement of the right intervertebral foramen at D.10/11 and destruction of the pedicles at this level. The 10th and 11th right ribs are splayed apart. A, Lateral; B, Anteroposterior.

ON EXAMINATION.—A man of middle age in good general health with no *café-au-lait* patches or fibromata. His blood-pressure was 160/105, the cardiovascular system otherwise being normal. The chest was more prominent on the right side, posteriorly, with expansion impaired at the right base, and there was well-marked dullness over the right lower lobe with poor air entry. The liver dullness extended abnormally high in the axilla and 4 fingerbreadths below the costal margin. Examination of the spine revealed scoliosis in the dorsal region with a free and painless range of movement and without tenderness of the spinous processes.

Neurological Examination.—There was no abnormality of mental state, skull, cranial nerves, or upper limbs. He had a severe spastic paraparesis, more marked on the left with considerable reduction of power, particularly at the knees and ankles. With extended knees he could lift the left heel from the bed for a distance of 3 in. only and the right for 9 in. The effect on function was even more marked because of gross spasticity. There were periodic severe extensor spasms of both limbs, brisk tendon jerks, inexhaustible ankle clonus, and extensor plantar responses. He was unable to stand alone; the right knee tended to flex repeatedly and he had great difficulty in standing up. Sensory testing revealed impairment of light touch and pain below D.10 spinal segment and absent vibration and joint sense below this segmental level. The lower abdominal reflexes were absent and there was weakness of the lower abdominal muscles.

Investigations.—Examination of the urine revealed a slight trace of sugar. A sugar-tolerance curve confirmed mild diabetes. The erythrocyte sedimentation rate was 42 mm. in the first hour.

RADIOGRAPHY.—

Dorsal spine: There was a mild upper dorsal scoliosis convex to the right (*Fig.* 347). The appearances were thought to be virtually diagnostic of a dumb-bell neurofibroma at D.10-11 with a large extraspinal extension.

Chest: A huge round mass at the right base was seen on radiography of the chest (*Fig.* 348). The conclusion was drawn that despite its size the mass was best explained as a huge thoracic neurofibroma arising from the area already suggested by the radiographs of the spine and possessing a small intraspinal extension.

Barium swallow: The œsophagus was deviated to the left and anteriorly by the tumour.

In Dr. Nevin's opinion this patient had a dumb-bell neurofibroma or ganglioneuroma and he was more in favour of it being the latter. He felt that the remission of symptoms in 1939 was due to release of compression on the spinal cord.

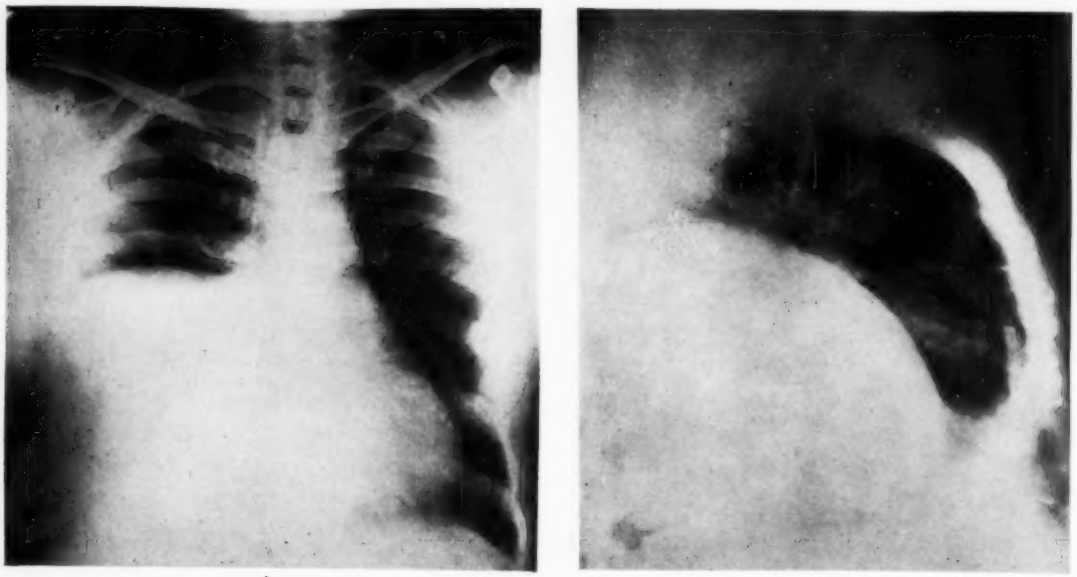
The neurosurgeon (Mr. Valentine Logue), who saw the patient later, also thought that the diagnosis was one of these tumours.

Special Investigations.—

Lumbar puncture: Clear cerebrospinal fluid under a pressure of 120 mm. was obtained. There was a moderate degree of manometric block. The fluid contained 50 mg. per cent of protein and was otherwise normal.

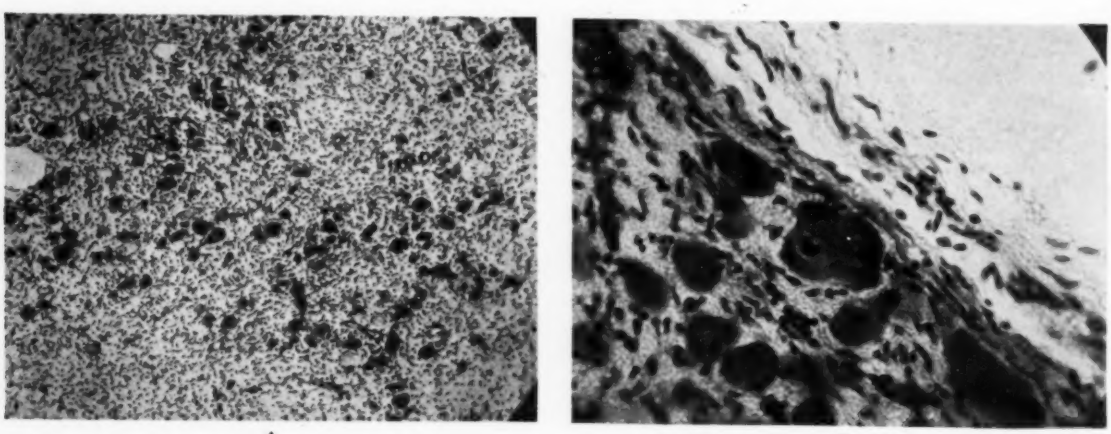
Lumbar myelography: This revealed a complete block to the flow of myodil at the lower border of D.11. The myodil column and cord impression were displaced to the left. These appearances were in favour of an extradural expanding lesion lying posteriorly and on the right lateral aspect, extending down to D.12 body on the right side.

With myelographic evidence of the lower level of the lesion being D.12, the plain radiograph changes suggesting tumour at D.10, and the clinical findings being in favour of the upper level at D.8, it seemed probable that the intraspinal component was a long lesion. In view of this, it was decided to operate in two stages, the spinal tumour being removed first.



A B

FIG. 348.—*Case* 1. A, Postero-anterior, and B, Lateral films of the chest showing a huge round mass at the right base. Superficially it resembles an elevated diaphragm, but the lateral view, an over penetrated postero-anterior film and screening, proves conclusively that it is intrathoracic and above the diaphragm.



A B

FIG. 349.—*Case* 1. A, Section of intraspinal component of tumour, showing numerous ganglion cells. (× 47.) B, Ganglion cells under higher magnification. (× 187.)

AT OPERATION (Mr. Valentine Logue).—A laminectomy from D.8–12 inclusive was carried out. The tumour filled the whole of the spinal canal posterior to the dura, made its entrance through a very large intervertebral foramen between D.10 and 11, and had tongues of growth 1 cm. in length, growing back again into the

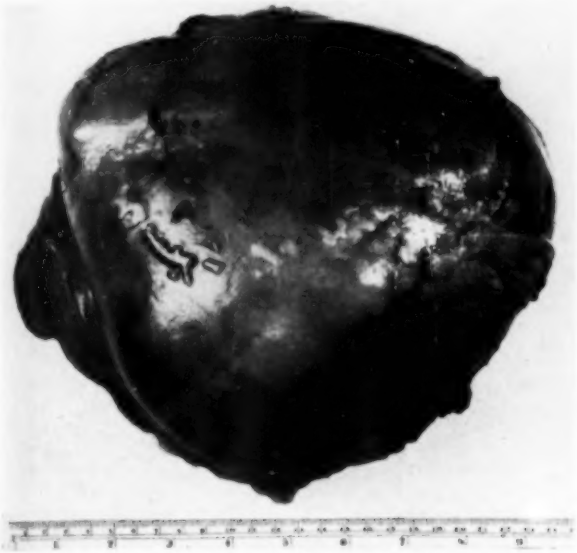


FIG. 350.—*Case* 1. Photograph of intrathoracic tumour.

right side of the chest through D.8-9 and 11-12 foramina. On the left, 4 similar but longer prolongations went out through the corresponding foramina.

The tumour weighed 36·5 g., and the histological report (Dr. C. E. Lumsden) was as follows: "It consists of ganglion cells of sympathetic type interspersed between the non-medullated nerve fibres and fibrous connective tissue. The nerve cells vary somewhat in size but the majority have one nucleus only. Double, treble and even quadruple nuclei are occasionally seen, the nuclei being fairly uniform in pattern. The lipochrome content varies very much but is considerable in some cells. Bielschowsky preparations show well developed axons in several of these cells. The appearances are those of ganglioneuroma." (*Fig.* 349.)

The patient was discharged to his home 5 weeks after the operation with considerable improvement in the control of his bladder and slight improvement in the power and spasticity of his lower limbs, but no significant change in the sensory loss. The diabetes, slight in degree, responded favourably to a low calorie diet, and insulin was not required.

Five months following the laminectomy, this patient was admitted to King's College Hospital under the care of Mr. W. P. Cleland for removal of the thoracic tumour. Examination at this time showed the chest to be barrel-shaped with poor expansion. There was bulging on the right side posteriorly up to the 5th spinous process with diminished air entry. His blood-pressure was 125/80 and he still showed a spastic paraplegia. The radiographic appearances of the chest mass have already been described.

SECOND OPERATION.—Through a right posterolateral thoracotomy, the large tumour, covered by mediastinal pleura, was exposed. The posterior ends of the 10th and 11th ribs were separated for a distance of 2½ in. They were splayed out and eroded and the 10th rib was dislocated upwards from its transverse process. The tumour had extended outside and around the 9th and 10th ribs. The 10th vertebral foramen was ½ in. in diameter and a process of the tumour was dissected from its orifice. The vertebral bodies were eroded, but the intervertebral disks were still prominent. The tumour was roughly oval in shape and 9 in. in its greatest diameter (*Fig.* 350). The diaphragm was depressed and flattened. Section of the tumour showed it to be a ganglioneuroma. It weighed 2830 g.

Apart from a loculated pleural effusion which cleared with repeated tapping, the patient made a good recovery.

The patient has been followed up and was last seen in April, 1956 (2½ years after the initial operation). There had been improvement throughout all this period to the extent that he was able to walk for distances up to a mile and could stand for quite long periods. He was free of pain and felt that bladder and bowel control were virtually normal. He was still using his mechanical chair to cover any distance, and when in unfamiliar surroundings preferred walking with a stick and tripod. He had had no symptoms referable to the thoracotomy. On examination, his legs were still spastic with brisk tendon reflexes and bilateral extensor responses. There had been some improvement in joint sense at the hips, but it was still absent at the toes and ankles and probably the knees also.

Case 2.—F. W. (M.V.H. 31095). Male, aged 35.

This patient was admitted in March, 1954, under the care of Dr. Helen Dimsdale. A single man, he was well until the age of 22. He was then in the army and a single 'black-out' led to a full neurological examination, von Recklinghausen's disease being diagnosed following biopsy of a subcutaneous nodule.

At the age of 23 there first appeared severe aches in the bones of the limbs and rather severe frontal headaches; also a little clumsiness of the hands and slight weakness of both lower limbs. After 6 months the aching pain disappeared, the headaches persisting. Ever since he had had occasional bouts of headache, usually frontal, but at times occipital. He had also experienced fleeting limb pains since that time.

However, 6-7 months preceding admission, the headaches had been persistent, and the clumsiness of his hands and weakness of his legs were more marked with

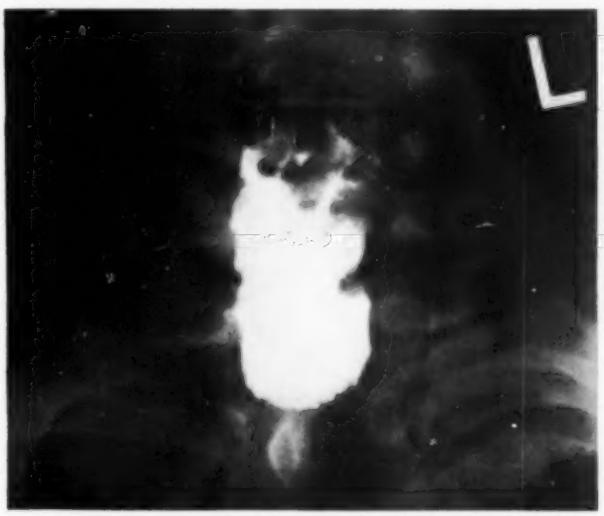


FIG. 351.—*Case* 2. Lumbar myelogram showing a block in the lower cervical region with multiple small rounded defects.

the most severe headaches. Three months prior to admission he had another episode of unconsciousness without convulsions. The pains in the extremities became more severe and, in addition, painful tender swellings of the joints appeared.

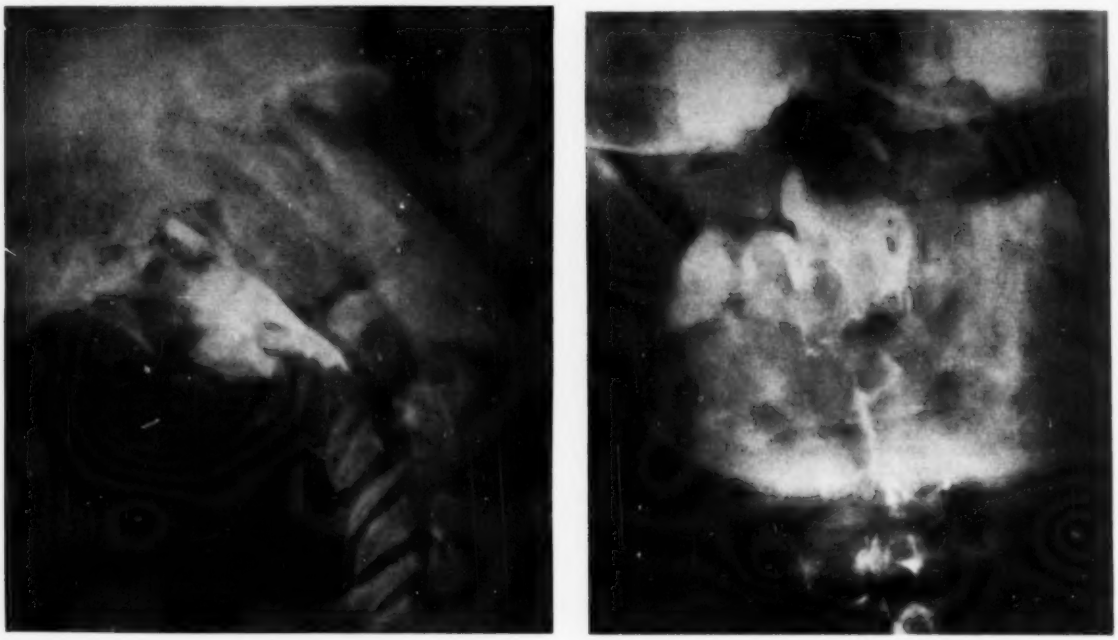
Direct inquiry revealed that for 6 weeks he had had considerable tingling and numbness in all four limbs. The lumps on his body were first noticed in 1939, and

had become more numerous since. They were increasing in size and were tender to touch. He had lost 1½ stone in weight over a period of 3–4 months.

Family history and previous medical history were not contributory.

On Examination.—The patient was a young man of pale complexion, of less than average height, with a fairly pronounced dorsal kyphosis and a compensating deterioration in intellectual capacity. His skull was slightly asymmetrical and he had a slight right upper motor neurone facial weakness and a Horner syndrome on the right side. His hearing was normal.

There was definite weakness and wasting of the shoulder-girdle muscles, but little loss of power in the distal limb segments. He had a moderately severe spastic paraparesis with bilateral extensor plantar



A B

Fig. 352.—*Case* 2. Cisternal myelogram showing upper cervical block with rounded defects. A, Lateral; B, Postero-anterior.

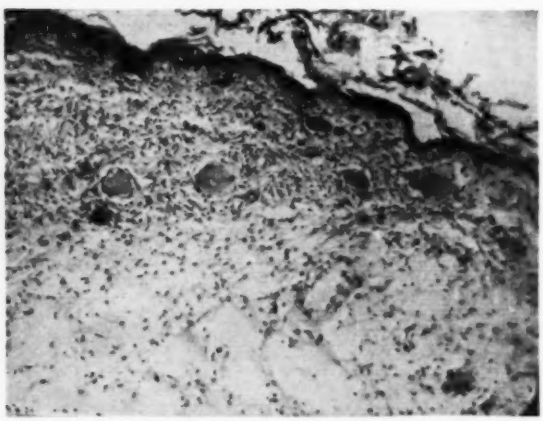


Fig. 353.—*Case* 2. Section of one of the tumours showing well-marked ganglion cells. (× 47.)

cervical lordosis. He had rather sparse bodily hair and an asymmetrical skull. He had multiple, widely distributed tender subcutaneous tumours of varying size. Some peripheral nerves had long chains of nodules on them and the brachial plexuses could be easily palpated because of swellings of their component parts, and were acutely tender. Most of the scalp nerves were palpable, some were visible, and all were tender. He had a few pedunculated neurofibromata, two large *café-au-lait* patches on the abdomen, and a rather large, cutaneous capillary angioma on the back of his neck. There was adequate evidence of rheumatoid arthritis.

Neurological Examination.—He was mentally slow and of poor intellect, but with no evidence of recent responses. The tone was also increased in the upper limbs with augmentation of the reflexes and positive Hoffmann responses. The abdominal reflexes were weak in the upper and absent in the lower quadrants. There was no significant disturbance of sensation or of sphincter function. He had a spastic gait.

Investigations.—The following yielded positive results: A radiograph of the right wrist revealed gross changes consistent with an infective arthritis. (The erythrocyte sedimentation rate in a previous hospital had been elevated.) An electro-encephalogram showed generalized dysrhythmia, suggestive of epilepsy.

The provisional diagnosis was spastic quadriparesis, due to intraspinal cervical neurofibroma in a patient with von Recklinghausen's disease and mild epilepsy. An associated condition was rheumatoid arthritis.

Special Investigations.—

Lumbar puncture: The cerbrospinal fluid was clear and under a pressure of 100 mm. There was a partial block on manometry and the fluid contained 200 mg. per cent of protein.

Lumbar myelogram: This showed a block in the lower cervical region, with multiple small rounded defects (*Fig.* 351).

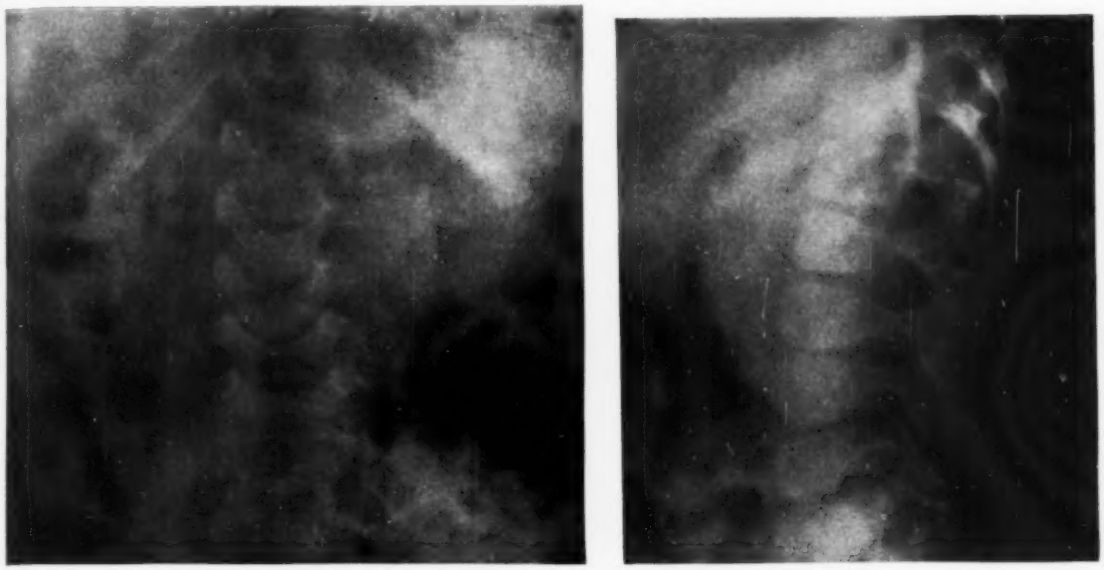
Cisternal myelogram: This revealed a number of rounded defects extending from C.2 to C.7 (*Fig.* 352).

First Operation (Mr. Valentine Logue).—An extensive cervical laminectomy from C.2 to C.6 was done. The spinal cord was displaced backwards by several tumours and on this occasion 3 were removed. The first tumour removed was from the right 5th root. It extended through the dura into the intervertebral foramen, being dumb-bell in form with respect to the dura, the extradural component extending into the foramen. It

measured 1½ cm. in length and 1 cm. in its other dimensions. The remaining tumours were also dumb-bell in shape. No attempt was made to remove the extradural portions of the tumours separately. Two further tumours at a higher level were left for subsequent laminectomy. The compression of the spinal cord was largely relieved at this time.

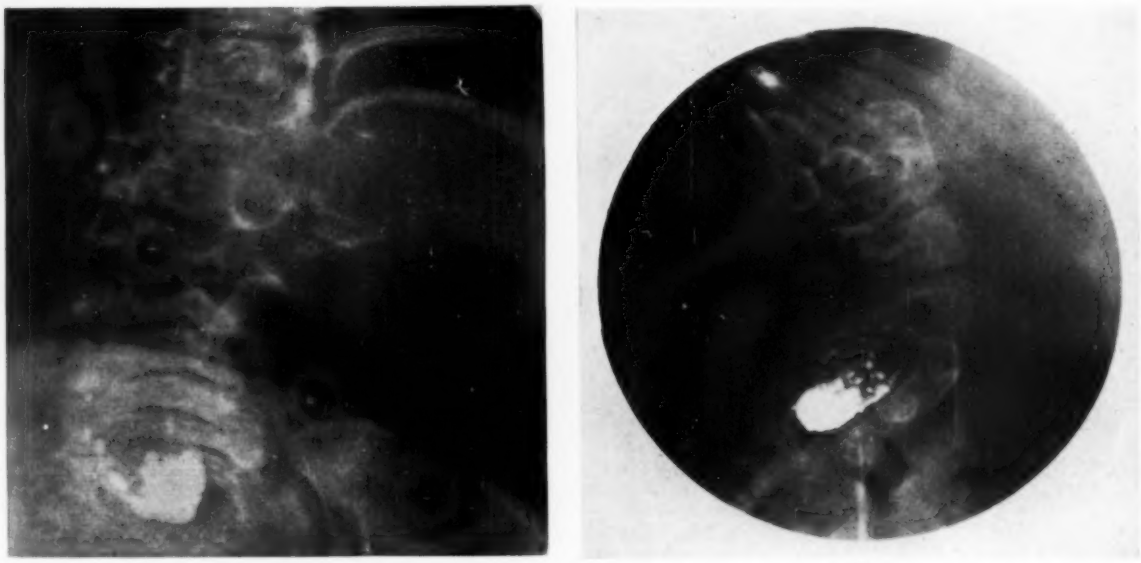
fact, are ganglioneuromata and the microscopical appearance recalls that seen in your recent case (*Case* 1) though the ganglion cells are less abundant in the present instance." (Dr. C. E. Lumsden.)

Following the operation the patient had retention of urine for a week and then normal micturition was resumed. There was improvement in power in the lower limbs and



A B

FIG. 354.—*Case* 3. Showing splaying of the pedicles in the lumbar region, erosion of the posterior aspect of L.1 and L.2, enlargement of the L.1/2 intervertebral foramen, and a soft-tissue mass on the left lateral aspect of the lumbar spine. A, Anteroposterior; B, Lateral.



A B

FIG. 355.—*Case* 3. Showing gross pedicle splaying, erosion of lumbar vertebræ from behind, enlargement of intervertebral foramina, and kyphosis at dorsilumbar junction. Some myodil is trapped below the tumour. A, Postero-anterior; B, Lateral.

Pathological examination showed each tumour to be covered by a firm fibrous capsule. Histologically (*Fig.* 353) each consisted of "fibrocytic interstitial perineurial fibroblasts of sensory and sympathetic ganglia but scattered throughout these are nerve ganglionic cells, some of which are almost normal in appearance though many show vacuolation and various stages of 'neuronophagia' by the satellite cells. Histologically there can be no doubt that these nodules are not neurofibromata but, in

lessening in the spasticity. Two weeks after the operation a further myelogram revealed the myodil to be in the lumbar region, immobile, and outlining 4 further small tumours.

SECOND OPERATION.—This was carried out 3 weeks after the first. The posterior arch of the atlas was removed and 3 further tumours excised, all anterior to the cord but arising from posterior roots. All three had extensions through the dural sac and the neck of each

was cut off flush with the dura. There were also a number of small (match-head size) tumours on the second cervical root on the left side. (A number of the lower cervical roots were seen to be similarly involved at the original operation.)

Pathologically, these tumours were essentially similar to those removed at the initial operation. The ganglionic cells were fairly abundant and of large mature form.

The neurological condition improved in the three weeks following operation, the shoulder movements

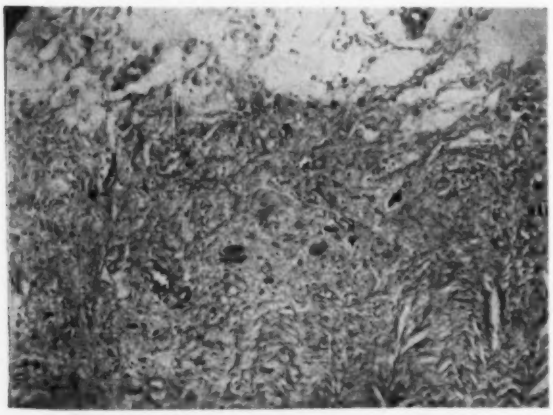
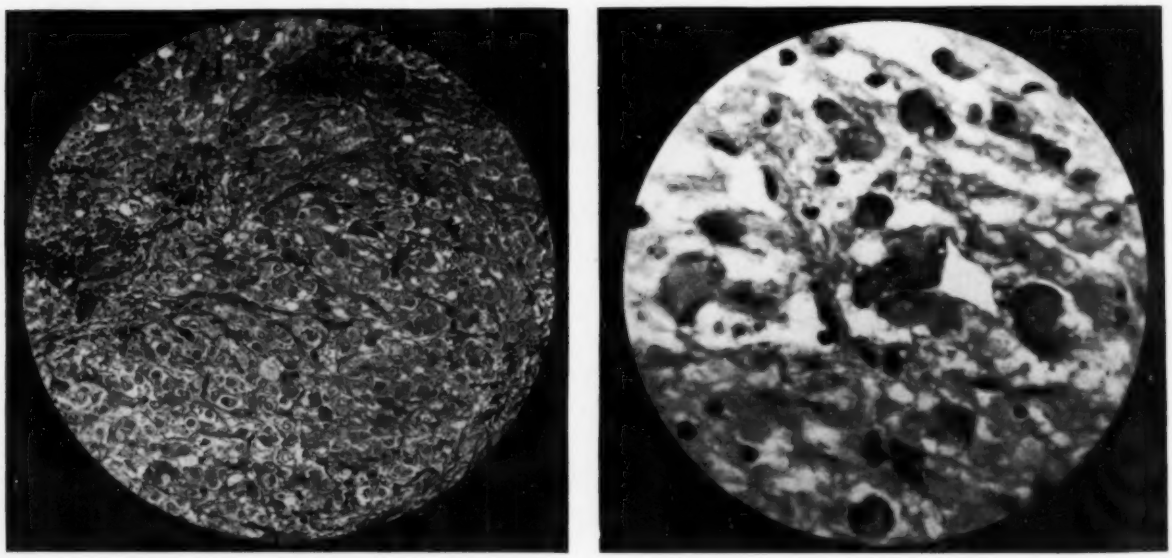


FIG. 356.—*Case* 3. Section of intraspinal portion of tumour showing ganglion cells. (× 47.)



A B

FIG. 357.—*Case* 3. A, Biopsy of abdominal tumour (1945). Area showing phæochromocytoma structure. B, Biopsy of abdominal tumour (1945). Area showing ganglioneuroma structure. (× 132.)

became much stronger, and the power in the lower limbs was four-fifths of normal. The tone diminished in all four limbs and his gait improved. There was, of course, sensory loss in the cervical dermatomes corresponding to the roots involved by the tumours. He developed a small meningocele for which operative repair was carried out 7 weeks after the second laminectomy.

The patient was seen for a follow-up examination 2½ years after the operation. During this period of time he had required subsequent admissions to hospital for treatment of rheumatism and headaches. However, his paraplegia had improved to a moderate degree. He was able to walk for periods of 15 minutes, to get about without the use of sticks, and to use the ordinary forms of public transport. At this time two of the subcutaneous abdominal tumours were removed and their nature as neurofibromata verified histologically.

Case 3.—A. M. H. Female, aged 7.

This patient was admitted to the Neurosurgical Unit of St. George's Hospital (at Atkinson Morley Hospital) in November, 1949.

When 11 months of age, her parents first noticed an inability to straighten her legs, and she was first seen at the Hospital for Sick Children, Great Ormond Street, at the age of 15 months with this complaint. In addition, profuse sweating, irritability, and hypotonia were noted. An abdominal mass was present at this time to the left of the spine and a radiograph revealed scoliosis in the lumbar region (*Fig.* 354). She was admitted to hospital 6 months later and at laparotomy the tumour was judged to be irremovable. A biopsy showed appearances typical of neuroblastoma. When seen again 18 months later there was no evidence of metastases.

In October, 1949, she was re-admitted to the Hospital for Sick Children with acute kyphoscoliosis and persisting paraplegia. The sections of the tumour were reviewed by the pathologist (Dr. Bodian) and a diagnosis of paraganglioma made. She was transferred to the Atkinson Morley Hospital under the care of Mr. Wylie McKissock.

ON EXAMINATION.—A child, small for her age, with normal cardiovascular and respiratory systems, and a blood-pressure of 110/70. She was able to sit up but not to walk, and had a marked kyphoscoliosis in the dorsilumbar region. There was a firm mass to the left of the vertebral column in the hypochondrium, fixed, about 3 in. in diameter, and visible from the back. The neurological examination revealed positive findings limited to the lower limbs. There was a marked flaccid paraplegia, the child being just able to lift each limb from the bed. The reflexes were absent and there was profuse sweating. The feet were deformed in a position of talipes equinovarus.

Investigations.—Routine laboratory examinations revealed normal findings; the spine radiograph showed marked changes, as indicated in *Fig.* 355. The appearances were interpreted as probably being due to a neurofibroma.

AT OPERATION (Mr. McKissock, December, 1949).—A large extradural smooth ovoid greyish-pink tumour

was exposed. The lumbar theca was pushed forward and to the right and was angulated through more than 90° at the kyphos. The upper portion of the tumour was removed at this operation. At a second operation one month later the lower part of the tumour was exposed, but due to close involvement with the rootlets of the cauda equina, only a partial removal was attempted. The communication between the intraspinal and intra-abdominal portions of the tumour was divided at the first operation.

PATHOLOGY (Dr. Bodian).—On histological examination the appearances were typical of a fully differentiated ganglioneuroma (*Fig.* 356). It is Dr. Bodian's opinion now—as indeed it was in 1949—that the biopsy obtained from the abdominal tumour in 1945 revealed appearances of a phæochromocytoma and not of a neuroblastoma. Parts of the tumour had a cellular structure typical of a ganglioneuroma. The photomicrographs (*Fig.* 357) show these varied appearances. The specimen of tumour removed at laminectomy in 1949 had the typical structure of a ganglioneuroma and no areas of phæochromocytoma were found in it.

The patient's immediate convalescence was satisfactory and there was slight return of motor power to the lower limbs. She has been followed up since that time. The paraplegia has persisted with associated urinary and fæcal incontinence. There has been no real change in the degree of kyphoscoliosis or in the size of the abdominal mass. She is now in a residential school for disabled children. She is able to sit unaided, but cannot stand or walk. There is no evidence of any intellectual impairment.

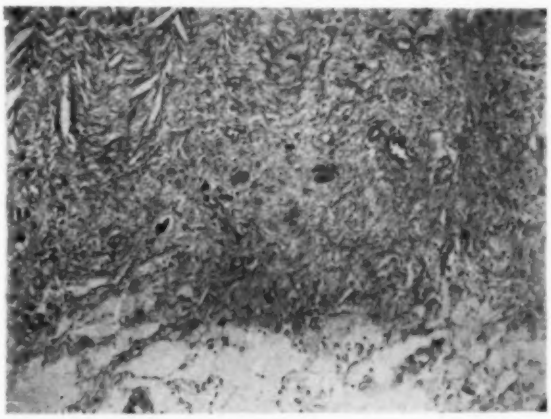
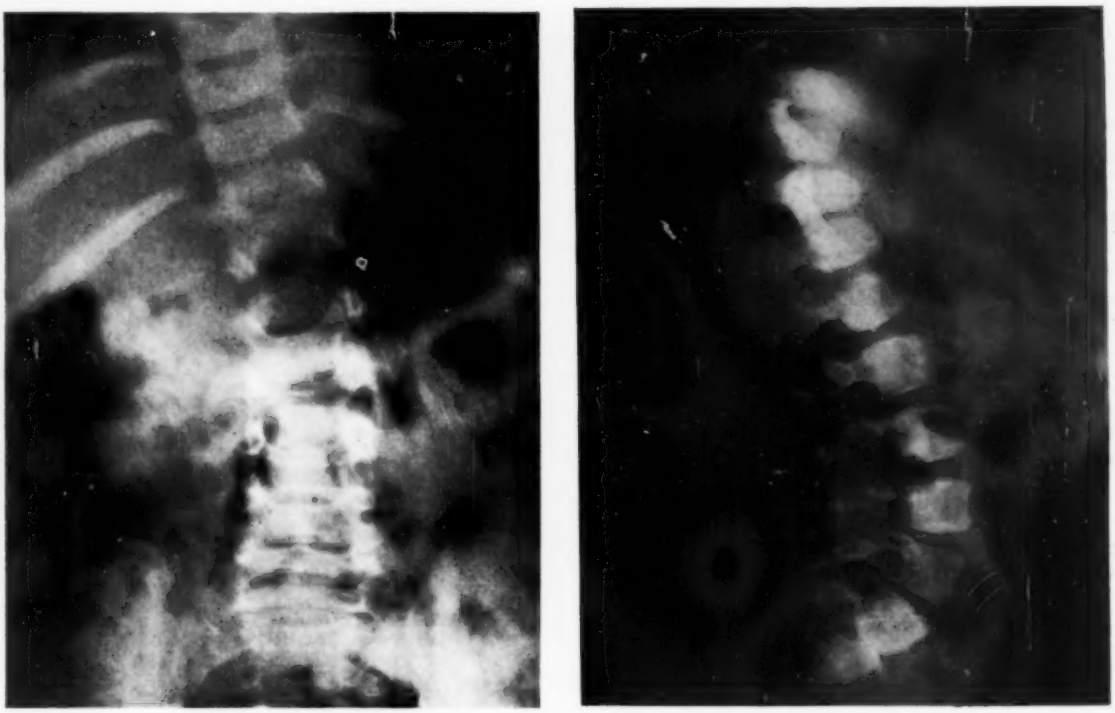


FIG. 358.—*Case* 4. Section of subscapular tumour showing ganglion cells. (× 47.)



A B

FIG. 359.—Dorsilumbar spine showing soft-tissue mass with calcification. Note also pedicle splaying. A, Antero-posterior; 3, Lateral.

Case 4.— L. C. Female, aged 2 years.

This patient was admitted to the Hill End Hospital (Neurosurgical Unit of St. Bartholomew's Hospital) in April, 1956, under the care of Mr. J. E. A. O'Connell.

She had never had full movement of her legs, and at the age of 11 weeks a lump was noticed in the left calf and at 5 months a subcutaneous swelling below the right scapula. At the age of 13 months the lump on the back was biopsied (Mr. F. E. Nash of Sydenham Hospital) and shown to be histologically a ganglioneuroma (*Fig.* 358). At no time was she able to walk and she did not develop sphincter control.

ON EXAMINATION.—She was a lively child who talked normally and could pull herself up if she had something to hold on to, but was quite unable to walk. Examination of the back revealed a swelling 3 in. by 2 in. just below the right scapula. There was no abnormality in the cranial nerves or upper limbs. There was spasticity of both lower limbs and diminution in their power. The gait (assisted) was typically spastic. The reflexes in the lower limbs were brisk and the plantar responses were both extensor.

Investigations.— Examination of the blood revealed normal findings. A radiograph of the abdomen (*Fig.* 359) revealed a paraspinal soft-tissue mass extending

from D.6 to L.4 and showing some calcification. There was a kyphoscoliosis centred at D.11 and abnormal development of the vertebræ below this level. In view of the calcification, a diagnosis of ganglioneuroma was favoured. A myelogram was carried out. This revealed complete obstruction at the level of L.2 and it outlined the lower end of a tumour which appeared to extend down to the level of L.3 on the right side (*Fig.* 360).

TREATMENT.—It was Mr. O'Connell's opinion that exploratory operation was inadvisable, and the patient

FIG. 360.—Lumbar myelogram showing complete obstruction of extradural type at L.2.

was referred to the care of Dr. I. G. Williams for X-ray treatment, 2000 r being given over a period of 55 days. A course of intramuscular vitamin B_{12} was also given, and intensive physiotherapy. There resulted considerable improvement in mobility, but at the time of her discharge on July 15, 1956, she was still unable to walk unaided.

DISCUSSION

The term 'dumb-bell' is used to describe tumours of characteristic shape. The most usual variety has a smaller intraspinal component with a larger paraspinal component joined by a constricted portion or neck occupying the intervertebral foramen. Eden (1941) described four types of dumb-bell spinal tumours :—

1. Tumours situated entirely within the spinal canal but being partly intra- and partly extradural.
2. Intra- and extradural tumours with a paravertebral extension.
3. Extradural and paravertebral tumours.
4. Foraminal and paravertebral tumours.

Heuer (1929) used the term 'hour-glass' tumours in describing these lesions. Entirely intraspinal tumours with intra- and extradural components were included in this term. It did not include malignant disease of the spine with intraspinal extension.

Heuer reviewed the various theories of the mode of formation of hour-glass tumours. Firstly, they may arise within the spine and grow outwards; secondly, they could arise outside the spine and grow inwards; thirdly, they might arise within the intervertebral foramen and grow in both directions; finally, the tumour may be already in situ before the development of the spine, much later, confers an hour-glass form upon it. Heuer subscribed to this last view, which was first proposed by Coenen (1927), and gives a delightful description of analogous tumours and cysts in other parts of the body.

The main features of the 15 cases available for study have been listed in *Table I*. This brings out several significant features.

Firstly, the anatomical localization of the lesions is predominantly dorsal or dorsilumbar. In only one patient was the cervical region involved and here the case was of a completely atypical nature. (This case will be further discussed below.) Dumb-bell neurofibromata, on the other hand—and these form the chief subject for differential diagnosis—can occur at any spinal level and are not infrequent in the cervical region.

Secondly, most of the cases showed an onset of symptoms in infancy (6 cases) or childhood (4 cases). The remaining 4 cases where dates of onset were given began at ages 17, 23, 26, and 63 respectively. This is in marked contrast to dumb-bell neurofibromata where the onset of symptoms is usually in adult life.

Thirdly, the tumours are benign, and provided that operative removal is undertaken before irreversible changes have been induced by the cord compression, the prognosis appears relatively good.

Ganglioneuromas are tumours of the sympathetic nervous system. They are not uncommon, and Stout (1947) collected 233 cases and added 10 more. He found that 60 per cent of cases occurred below the age of 20 years and that three-fifths of the patients were male. He pointed out that "ganglioneuromata originate from the sympathetic chain or rather from the cells of the anlage of the sympathetic chain and consequently are found in the cervical region, posterior mediastinum, posterior wall of abdomen, posterior aspect of the pelvis and the adrenal medulla". He quoted Wahl (1914) for the statement that they can also arise from elements within the central nervous system. He noted that occasional examples have been found in other sites (e.g., skin, tongue, and leg) presumably arising from ganglia.

It seems clear that the rare dumb-bell type of ganglioneuroma with which we are concerned in this paper is only likely to occur in association with the development of the paraspinal tumours. Nearly all the recorded cases have arisen from tumours of the sympathetic chain. Our *Case* 2 is an exception in that tumours had arisen within the spinal canal and extended outwards into intervertebral foramina. It is also exceptional in that multiple tumours were present, and in that the dumb-bell tumours occurred in the cervical region. We have not been able to find a previously reported case of cervical dumb-bell ganglioneuroma. The association with von Recklinghausen's disease is another unusual feature,

though it has previously been recorded by Stout (1947).

Pathology.—Bigler and Hoyne (1932) classify three basic types of sympathetic tumour, each varying in degree of malignancy and each derived from a different cell type :—

1. Neuroblastoma derived from neuroblasts and highly malignant.

2. Chromaffinomata or paraganglioneuroma derived from phæochromoblasts ; about 15 per cent of these become malignant.

3. Ganglioneuromata derived from the adult ganglion cell and benign. However, it is possible for intermediate types to occur and these form another group :—

4. Mixed tumours, thought to be derived from cell-nests ; these tumours may become malignant.

Stout in his review of 250 cases analysed 199 tumours from the histological point of view and sub-divided them into three groups :—

1. Fully differentiated (adult-cell type). This contained 146 cases, of which none became malignant.

2. Partially differentiated. This group contained 33 cases, of which 6 became malignant.

3. Composite, containing groups of completely undifferentiated cells. There were 20 cases in this group and 13 became malignant.

Stout's Group 1 would correspond with Group 3 of Bigler and Hoyne and his Groups 2 and 3 with their Group 4. Of the recorded dumb-bell sympathetic tumours nearly all appear to have been fully differentiated adult-cell type of ganglioneuromata (Stout's Group 1) and therefore benign. However, isolated cases of more malignant types have been recorded. A few, at some stage in the development, have probably been examples of the mixed group. In Cushing and Wolbach's patient (*Case* 5, *Table I*), for example, it seems reasonable to suppose that the tumour was initially of a malignant type (neuroblastoma) later undergoing maturation to the benign form. Cushing expressed the opinion that such maturation could occur. The same change possibly took place in our own *Case* 3 (*Case* 14 in *Table I*). However, both may have been examples of the second or partially differentiated group of Stout (*see Figs.* 356, 357). Rapp's patient (*Case* 3, *Table I*) may very well have had a tumour of mixed type as it was infiltrating muscles, although it was Rapp's personal opinion that the tumour was a ganglioneuroma. More malignant neurogenic tumours with dumb-bell characteristics have been recorded (Jacobs, 1955). In relation to these cases it is interesting that Kissane and Ackerman (1955) have suggested that neuroblastoma may occasionally undergo maturation following radiotherapy to the more benign ganglioneuroma, and have recorded a case. Whether these occasional histological problems are due to ' maturation ' is, however, of little practical importance. The majority of these rare dumb-bell sympathetic tumours will prove to be ' benign ganglioneuromata. Occasionally they may appear malignant and locally invasive in the early stages, but this appearance is not confirmed by their prolonged course and subsequent behaviour.

Intermittence of Symptoms and of ' Remissions '.—This feature was present in 2 of the previously reported patients—Cushing and Wolbach's (*Case* 5, *Table I*) and Eden's (*Case* 7, *Table I*) and in one of our present 4 patients (*Case* 13). A simple mechanical explanation is usually adduced to explain this, there being a variation in the size of the intraspinal component if some freedom of movement is possible through the enlarged intervertebral foramen. Recurrence does not occur in the true sense when complete removal of intra- and paraspinal portions of the tumour has been carried out. If the intraspinal portion only is removed then further extension through the enlarged intervertebral foramen from the paraspinal portion may occur, producing recurrence of symptoms. Similarly, removal of a paraspinal tumour without recognition of a foraminal or intraspinal extension may be followed by paraplegia or regrowth of the paraspinal mass. These remarks are also applicable to dumb-bell neurofibromata.

Paraplegia.—Eleven of the 15 patients were known to have had paraplegia. In common with other dumb-bell tumours this is one of the most important presenting features and forms a disability for which relatively urgent relief is required. In those patients with neurological disability the paraspinal component of the tumour may be relatively silent, in the clinical sense. In other cases it may give rise to symptoms, or be discovered during investigation for some other illness. However, when attention is drawn first to this portion of the tumour, the possibility of an intraspinal extension should be entertained and a careful neurological examination carried out.

Site of the Tumour with Reference to the Dura.—In 11 of the 15 patients the intraspinal portion of the tumour was extradural ; in 2 it was intradural, and in 2 the site was not clearly stated. With dumb-bell neurofibromata there is much more commonly an intradural component. This difference probably arises from the differing site of origin, ganglioneuroma arising from sympathetic ganglia or their primordia, and neurofibromata from spinal nerves or their roots.

Radiology.—Plain radiograph findings are described in 5 of the previous cases as well as in our own 4 cases. The extraspinal component of a dumb-bell ganglioneuroma, if it lies in the thoracic region, is readily visualized as a mediastinal mass. It may be hidden behind the cardiac shadow, however, but usually, unless it is very small, the mass is readily detectable. An abdominal mass is of course much less evident and may be palpable before it is evident on the plain radiographs. However, such a paraspinal abdominal mass may be quite easily visible (*see Figs.* 354, 359). With a mediastinal mass the differential diagnosis is usually from a dumb-bell neurofibroma, and pre-operative differentiation is hardly possible unless there is evidence of calcification. The age of the patient, as has already been noted, is a factor which may suggest the diagnosis. In our own *Case* 1 the extraspinal intrathoracic mass was so huge and low in position that it had previously been mistaken elsewhere for an enlarged liver, but such a mistake is really only feasible in an isolated exceptional case of this type. Calcification was present in our *Case* 4, and is also described in *Case* 9 (Sames, 1950). It is uncommon, but when it occurs is a point favouring

Table I.—DETAILS OF THE FIFTEEN RECORDED CASES OF DUMB-BELL GANGLIONEUROMATA OF THE SPINE

CASE	AUTHORS	AGE	LENGTH OF HISTORY	SEX	SYMPTOMS AND SIGNS	RADIOGRAPHY	OPERATION	HISTOLOGY	FOLLOW-UP	LEVEL	SIDE
1	Loretz (1870)	(years) 35	—	F.	No clinical history. Tumour size of hen's egg found at autopsy	—	—	Ganglioneuroma	—	D.2/2	—
2	Busse (1898)	4	4 yr.	M.	Paraparesis. Sphincter symptoms. Mass left hypochondrium	—	Abdominal mass only removed	Ganglioneuroma	Some improvement	Dorsilumbar	Left
3	Rapp (1913)	4	3 yr.	M.	Paraplegia. Sensory loss. Decubitus ulcers. Mass right lumbar region	—	Abdominal removal + spinal tumour	Ganglioneuroma (mixed tumour)	Not stated	Lumbar	Right
4	Stout (1924)	2½	2 yr.	M.	Paraplegia. Sphincter symptoms	Mass right upper mediastinum	Laminectomy	Ganglioneuroma	Not stated	Cervicodorsal	Right
5	Cushing and Wolbach (1927)	12	10 yr.	M.	Paravertebral swelling. Paraplegia	—	1. Exploration 2. Laminectomy	1. Neuroblastoma 2. Ganglioneuroma	Not stated	Thoracic	—
6	Naffziger and Brown (1933)	7	? 3 yr.	M.	Acute paraplegia. Mass right back. Sensory level D.8	Soft tissue mass D.5/D.10. ? inflammatory	1. Laminectomy 2. Laminectomy	1. Hæmangio-endothelioma 2. Ganglioneuroma	Died 3 mth. after second operation	D.3–7	Right
7	Eden (1941)	23	18 yr.	M.	Paraparesis. Sphincter symptoms for 3 weeks. Level D.5	1. At 5 yr. old, paraspinal mass in chest 2. At 23 yr., large mass left upper mediastinum. Enlarged dorsal foramina	Laminectomy	Ganglioneuroma	Died at operation	Dorsal	Left
8	Paterson and Pilcher (1941)	4	3 yr.	M.	No neurological signs. 'Asthma' since age of one	Mass right posterior mediastinum. Erosion 4th rib. Widened 4th intercostal space	Thoracotomy. Small extension into I/V. foramen removed	Ganglioneuroma	—	Dorsal	Right
9	Sames (1950)	3	2 mth.	F.	Paraplegia—level D.4	Right posterior mediastinal mass, calcified	Laminectomy. (Tumour anterior to and across cord)	Ganglioneuroma	5 yr. later some spasticity	D.3, 4, 5	Right
10	Rogers and Keogh (1950)	26	6 wk.	F.	Pain left arm and shoulder. No signs	Mass left upper mediastinum	Thoracotomy (extension into D.3)	Ganglioneuroma	—	D.2	Left
11	House and Bahrend (1953)	64	1 yr.	M.	Cough, dyspnœa. No neurological signs	Mass left posterior mediastinum. Enlarged intervertebral foramina. Myelogram: block D.10	Thoracotomy	Ganglioneuroma	Recovery	D.10	Left
12	Shephard and Sutton (1958) *Case* 1	53	35 yr.	M.	Paraparesis. Pain. Bladder symptoms. Sensory signs	Mass right chest. Enlarged foramina. Erosion vertebræ and ribs. Myelogram block	Laminectomy Thoracotomy	Ganglioneuroma	Improved	D.10	Right
13	*Case* 2	35	12 yr.	M.	Headache. Blackouts. Von Recklinghausen's disease. Spastic paraplegia. Rheumatoid arthritis	Myelogram: multiple rounded tumours with block	Laminectomy	Ganglioneuroma	Improved	C.2–7	—
14	*Case* 3	7	6 yr.	F.	Paraplegia: mass in left abdomen	Pedicle splaying. Enlarged foramina. Left lumbar mass. Myelogram: block	1. Laparotomy 2. Laminectomy	1. Mixed tumour (phæochromocytoma + ganglioneuroma) 2. Ganglioneuroma	Improved	L.1–4	Left
15	*Case* 4	2	18 mth.	F.	Paraplegia. Lump in back	Pedicle splaying. Soft-tissue mass with calcification. Myelogram: block	Biopsy	Ganglioneuroma	Improved after radiotherapy	D.—L.	Right

ganglioneuroma as against neurofibroma. *Case* 6 described by Naffziger and Brown showed a soft-tissue paraspinal mass, which was thought to be of possible inflammatory origin. The absence of any gross disk destruction should usually prevent such a misdiagnosis, as should the well-rounded nature of the tumour mass.

Enlargement of intervertebral foramina was present in 3 of our cases and in several of the other cases described, and is readily detectable on the radiographs. Where the tumour extends along the spinal canal, pedicle splaying and pedicle erosion may also be seen.

Myelography will adequately outline the level of the spinal block, and if necessary the upper and lower limits of the tumour can be shown by myodil injected both from above and from below. Usually the myelogram will show the characteristic appearances of an extramedullary, extradural expanding lesion.

SUMMARY

The literature on dumb-bell ganglioneuromata is reviewed and 4 new cases are added to the 11 previously recorded. No special search was made for our own cases and we feel that such tumours probably occur more frequently than the literature would suggest. The histological, clinical, and radiological aspects are discussed.

The variation in histological structure of the neurogenic tumours and the tendency towards maturation to the benign form (ganglioneuroma) are interesting features. The implication is that a more radical policy towards treatment may be appropriate in some cases, especially in children, before irrecoverable neurological damage is produced.

Points of clinical and radiological importance which may help towards a pre-operative diagnosis of the lesion are stressed.

Acknowledgements.—Our thanks are due to Dr. Helen Dimsdale, Dr. Samuel Nevin, and Mr. Valentine Logue for their permission to publish the records of the Maida Vale Hospital cases, and to Mr. W. P. Cleland for providing us with a description of the thoracotomy in *Case* 1 and with a photograph of the operation specimen. We are grateful to Mr. Wylie McKissock and Mr. J. E. A. O'Connell for allowing us access to their notes on *Cases* 3 and 4 and for permission to publish. We are indebted to Dr. W. H. McMenemey for allowing us full use of the histological material in *Cases* 1 and 2, and to his department for the photomicrographs; and to Dr. M. Bodian for the pathological report in *Case* 3, and for the photomicrographs (*Fig.* 357).

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INTRATHECAL SPINAL METASTASES*
(RARE TUMOURS)

BY LAMBERT ROGERS AND GORDON HEARD, CARDIFF

WHEN the spinal cord is involved by malignant metastasis the deposit of tumour almost always lies in the epidural space and compresses the cord through the dura mater, which it rarely invades.

While cerebral metastases are relatively common, spinal intradural metastases are rare and isolated ones exceedingly rare. In suggesting a reason for this rarity of spinal intrathecal and intramedullary metastases we here record a case and refer to the few which have so far been reported.

The nature of a particular tumour causing compression of the spinal cord cannot always be determined before operation. The history and clinical findings or the presence of a malignant lesion elsewhere in the body or of a history of one having been removed may lead to a suspicion of a metastasis. Occasionally, however, a metastasis may at operation resemble a benign tumour and be identified only on microscopical examination after its removal, as in the case here reported.

CASE REPORT

An adrenal metastatic intrathecal spinal tumour simulating a meningioma in a man aged 54.

An insurance manager, aged 54, referred by Dr. Grahame Jones, of Griffithstown, complained of pain in the back and around the right side of the chest below the shoulder-blade. This had been present for four to five months, with weakness and stiffness of both legs for the past two months. More recently his feet had felt numb and he had been unable to carry on his work.

He was apparently a healthy man, with no evidence of malignant disease or of ever having had a malignant tumour. The cranial nerves and upper limbs were normal. In the legs, tone was increased and both plantar reflexes were extensor. The lower abdominal

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reflexes were absent. All forms of sensation were impaired below the level of the sixth thoracic dermatome, pin-prick and temperature sense more so on the left side, vibration and position sense more on the right.

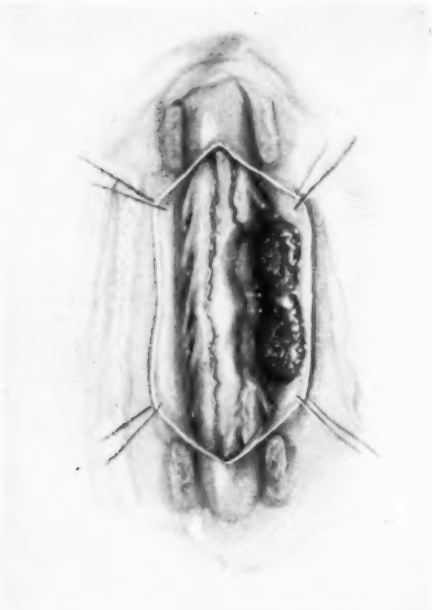


FIG. 361.—Appearance of tumour as exposed at operation

Radiographs of the vertebral column and of the chest were normal. His hæmoglobin was 96 per cent and the serum acid phosphatase content was 0·8 unit. Lumbar puncture yielded clear fluid with a pressure of 110 mm. On jugular compression there was a free rise but a somewhat delayed fall. The protein content was 280 mg. per 100 c.c. Myelography demonstrated an incomplete hold-up and filling defect at the level of the fourth thoracic vertebra and to the right.

At laminectomy a bi-lobed intrathecal tumour was disclosed projecting from the inner surface of the dura mater, posterior to the cord and on its right side (*Fig.* 361). The tumour was removed together with the area of dura from which it was arising, as it closely resembled and was thought to be a meningioma. The spinal cord exhibited indentations in which the tumour had lain but otherwise appeared normal.

On microscopical examination the tumour was regarded as a secondary carcinoma (*Fig.* 362). A renewed search failed to produce any evidence of a primary tumour.

The immediate post-operative course was satisfactory, but within a few weeks his general condition began to deteriorate; he developed a more profound paraplegia, and three and a half months after operation died from bronchopneumonia.

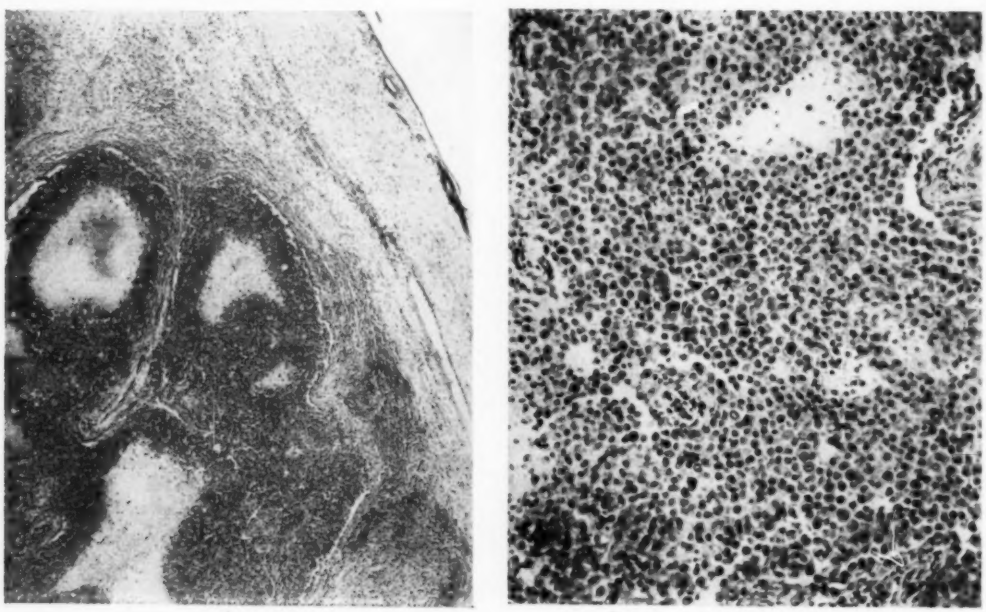
AT AUTOPSY.—Both adrenals were found to be the site of tumour, that on the left being entirely replaced by spheroidal-cell carcinoma measuring 6 × 4 × 2 cm. This was regarded as the primary lesion (*Fig.* 363). There was a similar deposit in the right adrenal.

The spinal cord at the level of the laminectomy was soft and on microscopical examination was found to be invaded by tumour (*Fig.* 364).

No other deposits of tumour were evident; the lungs showed widespread bronchopneumonia.

COMMENTS

Of a series of 111 spinal tumours operated upon in this Unit during the past thirty years, 56 were malignant and 17 of these metastatic. Only one of the metastatic tumours was intrathecal (the case here reported). Another patient, a married woman aged 51, with generalized Hodgkin's disease in addition to extradural deposits, had also a subpial mass of lymphadenoma.



A B

FIG. 362.—Sections of intrathecal metastatic adrenal tumour adherent to inner surface of dura. A, × 25·5; B, × 92.

Of 53 cases of metastatic spinal neoplasms investigated at the Philadelphia General Hospital by Wilson and Rupp (1947) only 2 were intramedullary, one being of adrenal origin, the other a deposit of lymphadenoma. Thus the incidence in

the Philadelphian cases resembles that in our series.

Few instances of intrathecal or intramedullary spinal metastases have been reported in the literature and these mostly single cases in which metastases have been widespread.

Amyot and Laurin (1939) reported a metastasis in the lower thoracic part of the spinal cord along with secondary deposits in the brain of a woman whose breast had been removed four years previously. In an extensive review of the French literature of the preceding ten years they could find no reports of metastases in the spinal cord. Single cases have been reported by Spencer (1951), Rasch, Rosenbaum, and Heimburger (1953), and Cantor and Stein (1952) in men aged respectively 56, 45, and 68, the primary lesion being in a bronchus in each case and the metastasis to the cord, part of a general dissemination with metastases elsewhere. In reporting their case Cantor and Stein were able to find only 13 other verified cases. Three of these arose from primary tumours in the breast, 3 in the kidney, 3 in the lung, 1 in the thyroid, 1 in the adrenal medulla, and 1 was a deposit of lymphadenoma, while the origin of the primary tumour in the remaining case is not stated.

Several writers have commented on the fact that macroscopical examination of the cord may not reveal an intramedullary metastasis, and in Cantor and Stein's case there was no spinal block present and the protein content of the lumbar fluid was only 71 mg. per 100 c.c.

The Reason for the Rarity of Spinal Intrathecal Metastases.—It will be seen that in contradistinction to the relative frequency of cerebral metastases from certain types of malignant disease, particularly carcinoma of the lung, the rarity of secondary deposits within the spinal theca is notable, though it may be that these occur slightly more often than is always realized, since they may not be detectable on naked-eye inspection of the cord. This variation between the incidence in the brain and in the cord may in part be due to the relative difference in the mass of the brain compared with the spinal cord, but may also depend on the vascular arrangements, which may be expected to favour the passage of neoplastic emboli to the brain rather than to the cord. The blood-supply to the cord, derived from small, late arterial divisions, presents a more tortuous route to tumour emboli than the direct passage to the brain through the carotid or vertebral arteries, while Batson (1940) and others have drawn attention to the small size of the veins of the cord as compared with the large vessels connecting the vertebral veins with the intracranial dural sinuses.

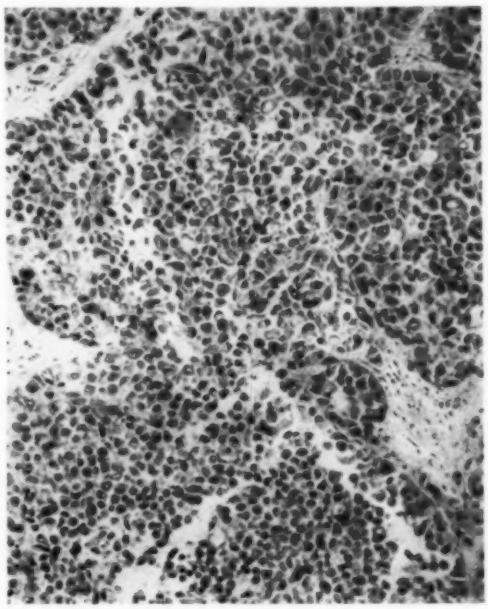
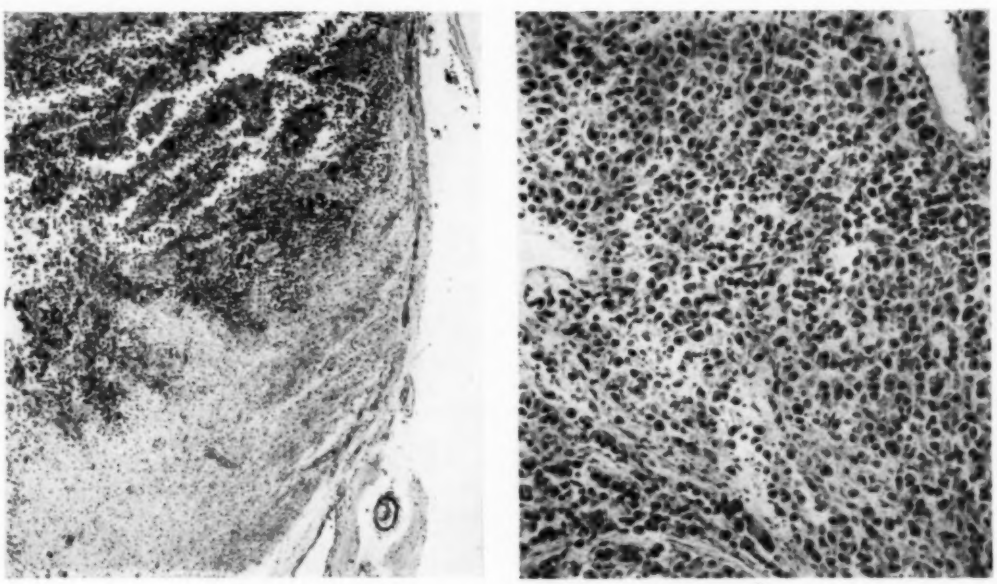


FIG. 363.—Primary tumour in adrenal. (× 92.)



A B

FIG. 364.—Invasion of spinal cord by tumour. A, × 25·5; B, × 92.

SUMMARY AND CONCLUSION

Metastatic tumours within the spinal dura mater or in the cord are rare lesions. Although the cord is not infrequently affected by metastatic tumours, these are nearly always extradural, and compress it from the epidural space for which they have a predilection. The spinal dura is remarkably protective, being very rarely either invaded or penetrated by malignant tumours lying outside it.

It is suggested that the relative difference in the incidence of intracerebral and intramedullary metastases is due to the difference in mass between brain and spinal cord and of the vascular arrangements which facilitate the passage of neoplastic emboli to the brain rather than the cord.

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HERNIA REPAIR USING NYLON TRICOT IMPLANT

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INTRODUCTION

IT is generally agreed that the cure of large inguinal herniæ, whether direct or indirect, and also the cure of sliding or recurrent inguinal herniæ are not easy and, as a result, it was decided to treat ruptures of this type with a nylon tricot implant either as reinforcement to a repair or as the sole means of repair. (*Lancet*, 1951.)

Nylon tricot can also be used in repair of large umbilical, incisional, and some diaphragmatic herniæ. It can also be used to advantage in repair of large defects of the abdominal wall after wide excisions in cases of malignancy.

Many materials have been used, but none has been found so uniformly satisfactory for a variety of reasons.

THE MATERIAL

Nylon tricot has a special open weave and is very thin and strong; it has a two-way stretch which makes fitting and suturing easy (and its support very effective); it does not fray. Granulation tissue can grow through the open mesh easily and serum is unlikely to pocket on its deep aspect. It is inert, and although cases of sensitivity to nylon are reported, no such case has been noted in this series. It is sterilized by boiling or autoclaving. It is cheap, readily available for use in any size or shape, and easily obtainable from any ladies' stores.

USE OF IMPLANT IN REPAIR OF LARGE HERNIÆ WITH POOR TISSUES

1. Inguinal Herniæ.—

Operation.—The hernial sac is ligated and removed and the spermatic cord mobilized. The cremasteric vessels are ligated and divided and repair of the transversalis fascia performed around the internal abdominal ring if possible. If it is now possible to approximate the conjoined tendon to the inguinal ligament with the help of the Tanner slide without tension (1942) this is then done, using interrupted non-absorbable suture material. In the event of this procedure being impossible the repair is left entirely to the nylon tricot implant. The nylon tricot implant is then inserted as follows.

Method of Insertion of Implant.—The spermatic cord is retracted. The nylon tricot implant, usually of size 5 in. long by 3 in. wide, held at each corner by Spencer Wells forceps, is laid lengthwise over the posterior wall of the inguinal canal and sutured at its medial and inferior corner to the strong aponeurosis overlying the pubic spine at the extreme inner end of the inguinal canal. A continuous suture is then used to fix the lower margin of the implant to the inguinal ligament, taking bites at intervals of ¼ in., and keeping the implant fairly taut, to a point about 1 in. lateral to the internal abdominal ring. Another continuous suture is now inserted from the pubic spine, fixing the upper edge of the implant to the upper aspect of the internal oblique aponeurosis to cover the deficiency of the Tanner slide, and continued to about 1 in. lateral to the internal abdominal ring.

A longitudinal slit is made from the outer narrow side of the implant about ½ in. vertically from the inguinal ligament to make two limbs fitting snugly, but not tightly, around the internal abdominal ring. The implant can be trimmed as necessary during this procedure and should be quite taut (*Fig.* 365). The wound is now closed in layers without drainage.

The needle usually used is a Kelly No. 4 intestinal half-circle, and suture material braided nylon, size 6 lb.

A course of penicillin, in dose of 1 mega unit daily for five days, is given in the post-operative period.

Follow-up Results.—Eighty-three hernia repairs have been performed in 73 patients during the past four years with no mortality. In 31 of these repairs herniotomy with a nylon tricot implant only was used. In the remaining 52 repairs the nylon tricot was used to support a Bassini repair. Fifty-eight cases have been followed up for one year or more; one hernia only has recurred, i.e., one-year recurrence rate 1·7 per cent. This recurrence occurred in a case where herniotomy and nylon tricot implant alone

was performed owing to a deficiency of suitable tissues.

Eight recurrent herniæ, of which two had recurred twice, have been followed up for one year. None of these has recurred.

Infection: Three cases of infection occurred, i.e., 3·6 per cent, in the very early cases of this series, but no recurrence occurred in these cases and all settled after widely opening the wound.

Incisional Hernia.—Nylon tricot implant has been used as a support for any repair or as the sole means of achieving repair.

Anterior Abdominal Wall Defects.—After wide excision of the anterior abdominal wall for desmoid tumour, sarcoma, or epithelioma, a wide deficiency will often result. Here an implant to repair the deficiency is invaluable and nylon tricot is excellent for this purpose.

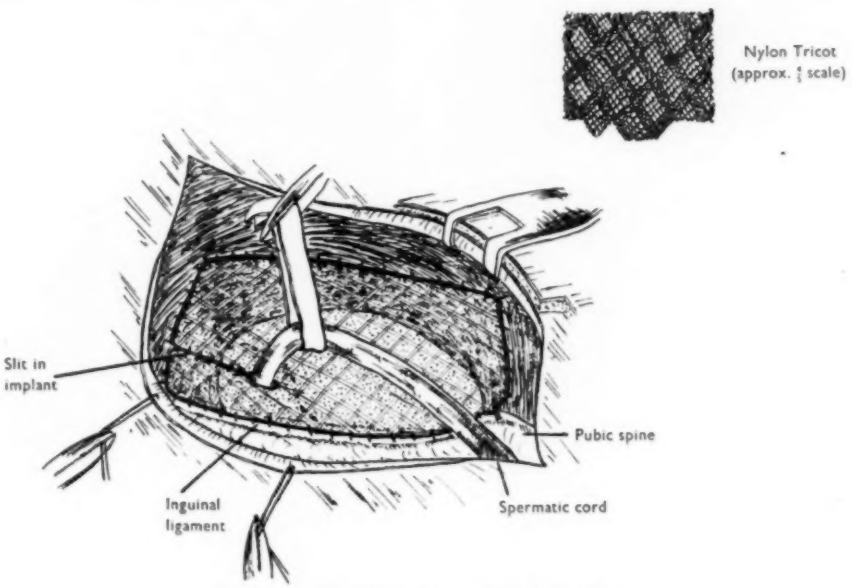

FIG. 365.—Diagram showing nylon tricot in situ.

2. Application of Use of Nylon Tricot in other Herniæ or Anterior Abdominal Wall Deficiency.—

Umbilical Herniæ.—Large wide-necked umbilical herniæ are very liable to recur and the use of this implant to strengthen a Mayo repair as an overlay, or actually to cover the deficiency in the linea alba without attempting to close it, is occasionally invaluable.

Hiatus Hernia.—When a large deficiency is present the musculo-aponeurotic tissues of the right crus, forming the margins of the œsophageal hiatus, are stretched and repair by suture of these may be difficult. In order to repair this deficiency effectively, sutures may so tighten the lower end of the opening that obstruction of the œsophagus may result.

In the first case treated by tricot implant the musculo-aponeurotic tissues around the œsophageal hiatus, and particularly behind the œsophagus between it and the aorta, were so deficient that the aortic and œsophageal openings appeared to be as one. A transverse strap-like implant of nylon tricot was used to repair this deficiency, which would otherwise have been very difficult to repair.

In later cases the nylon tricot implant has been used, after removal of the hernial sac, either as a reinforcement of a repair by sutures or as the sole means of repair. The approach used is either thoracic or abdominal, preferably the latter. A number of cases operated on in this way have so far been uniformly satisfactory. (*Figs.* 366, 367.)

DISCUSSION

The use of foreign materials in hernia repair has a definite although restricted value.

It is well known that none of these materials is perfect, but as far as can be judged, nylon tricot has no serious drawback.

The most frequent trouble met with in all these is infection. In this series the three cases of infection occurred early but no case of infection has been recorded for over three and a half years. In no case so affected was there a recurrence of the hernia. If infection does not settle with antibiotic treatment and is prolonged, re-opening of the wound and traction on the implant are usually sufficient for its removal and cure of infection.

Darns.—A large number of materials used in this manner for repair of hernia tend to result in a good deal of approximation of structures unable to withstand the slightest pull for long.

Darns such as silk floss (Maingot, 1941), Chinese twist silk used as a darn for the internal ring (Ogilvie, 1937), nylon (Moloney, Gill, and Barclay, 1948), and fascia lata strips (Gallie, 1921) besides all having this tendency to approximate the structures, as above, require often the use of a large needle with resultant fraying of the inguinal ligament. They also leave weak spots between the strands, or tight bands around the internal ring, which constrict the spermatic cord.

Implants.—Tantalum gauze (Cokkinis and Bromwich, 1954; Burnell, 1953), filigrees (McGavin,

1909), plastic plates (Thompson, 1948), and polyvinyl sponge (Schofield 1955) are difficult to handle, painful to the patient, and have a great tendency to disintegrate with bending. Whole-thickness skin-grafts (Mair, 1945) have a high incidence of dermoid cyst formation.

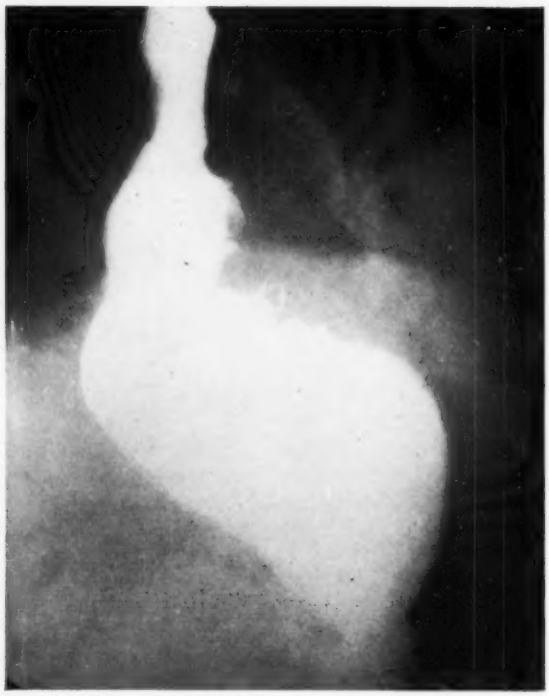

FIG. 366.—Barium swallow. Hiatus hernia with short œsophagus before repair.

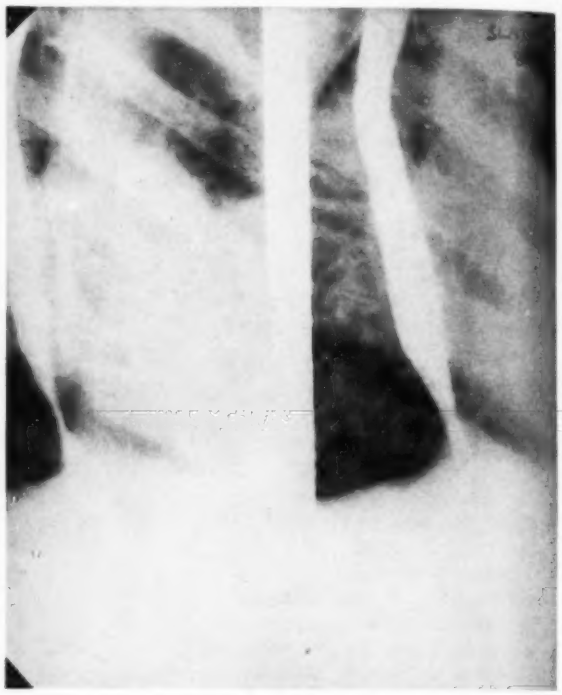

FIG. 367.—Barium swallow. Hiatus hernia repair after using nylon tricot implant.

Recurrence in cases of implants usually occurs at some point around the edge of the implant. It is, therefore, wise to apply the implant widely overlapping the extent of deficiency to be repaired.

The principle of the use of the nylon tricot implant is firstly to act as reinforcement to a repair of poor tissues which require a support, and secondly, as an actual replacement when the implant becomes surrounded and infiltrated by fibrous tissue, but itself remains intact.

SUMMARY

A method of repair of large and recurrent inguinal herniæ and other types of herniæ by use of nylon tricot implant is described. Eighty-three cases of inguinal hernia have been so operated upon and a number of cases of hiatus hernia, and isolated cases of other herniæ and deficiencies.

The mortality was nil. The one-year recurrence rate was 1·7 per cent. Three cases of infection (3·6 per cent) occurred in the early cases of the series.

The suitability and ease of use and sterilization are noted.

I wish to acknowledge the help of Mr. S. V. Unsworth, Senior Consultant Surgeon to the Liverpool Stanley Hospital, who first conceived this method of repair over six years ago, in the preparation of this paper; in particular for allowing me to publish an account of his repair of hiatus hernia.

I wish to thank Doctor D. Jacobs for *Fig.* 365.

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THE TREATMENT OF COMPLETE PROLAPSE OF THE RECTUM BY THE ROSCOE GRAHAM OPERATION

By J. C. GOLIGHER

FROM THE GENERAL INFIRMARY AT LEEDS

In 1933 the late Mr. Ernest Miles reported his experiences in the treatment of complete prolapse of the rectum by the operation of amputation, or rectosigmoidectomy, first described by Mikulicz in 1888. He had operated on 31 cases by this method with only one operative death and one recurrence, the latter having taken place more than five years after the operation. Subsequent reports on this procedure by Abel (1933) and Gabriel (1945) were also distinctly favourable, and as a consequence it became the standard treatment adopted by many surgeons in this country for complete prolapse of the rectum.

In 1949, however, Hughes conducted a comprehensive analysis of the results of rectosigmoidectomy for prolapse in 150 cases treated by this method at St. Mark's Hospital between 1931 and 1947 inclusive. He confirmed the great safety of the operation, for there were no operative deaths. Of these cases 108 were successfully traced, practically all of them attending for re-examination. No less than 65, or 60 per cent, of these were found to have developed a recurrence of their prolapse. In addition, Hughes elicited the disquieting fact that 50, or 46 per cent, of the traced patients were incontinent, and Goligher and Hughes (1951) suggested that this was largely a sensory incontinence due to excision of the sensitive ano-rectal mucosa by the rectosigmoidectomy. These findings showed beyond doubt that rectosigmoidectomy was an unreliable operation for rectal prolapse. Admittedly, the risk of impairment of rectal sensation with resulting incontinence can possibly be lessened by modifying the operative technique to ensure that a cuff of at least 1½ in. of ano-rectal mucosa is preserved in the anal remnant, but an operation that is followed by recurrence in over half the patients treated by it can scarcely be regarded as satisfactory.

Actually, when the theoretical basis of rectosigmoidectomy for prolapse is critically examined, it is not at all surprising that the operation should frequently fail, for its sole effect is to shorten the distal large bowel and render it taut. It does little or nothing to eliminate the main underlying causes of the condition, which are now generally held to be the following :—

1. The Abnormally Deep Recto-vaginal or Recto-vesical Pouch of Peritoneum.—This is a most striking and constant feature in all cases of complete rectal prolapse. The significance of the deep peritoneal pouch was early appreciated by Jeannel (1896), Quénu and Duval (1910), and especially by Moschcowitz (1912), who pointed out that complete rectal prolapse was really a form of sliding hernia, the pouch of Douglas being the hernial sac which presses the anterior rectal wall into the rectal lumen, and then through the anal canal to the exterior. This conception of the mechanism of prolapse (*Fig.* 368) has been strongly championed by Roscoe Graham (1942) in recent years.

2. The Lax and Atonic Condition of the Musculature of the Pelvic Floor and Anal Canal.—Nearly all cases of complete prolapse show a remarkably weak and atonic condition of the anal sphincters and levator ani muscles. It has always been a matter for dispute whether this state of affairs is the cause or an effect of the prolapse, but it has been observed often in very early cases of prolapse

FIG. 368.—Diagrammatic sagittal sections of the pelvis to demonstrate the anatomy of complete rectal prolapse conceived as a sliding hernia of the pouch of Douglas.

in which there hardly seems to have been time for the prolapse to have produced such secondary changes in the musculature.

3. Lack of the Normal Fixation of the Rectum to its Bed.—Jeannel (1896), Pemberton and Stalker (1939), and Muir (1955) have claimed that an abnormal mobility of the rectum in its bed is a factor in the causation of prolapse. Whether this is so or not, one principle employed in certain of the operations used for this condition is the promotion of adhesions between the rectum and its surroundings.

Rectosigmoidectomy can do little to rectify these various factors which are believed to predispose to the development of prolapse. If the pelvis is examined through the abdomen at the conclusion of a rectosigmoidectomy, as Thompson (1949) has pointed out, the abnormally deep pouch of Douglas is seen to persist virtually unchanged. The stretched condition of the pubo-rectalis muscles is also unaffected by this operation, and the advocates of the procedure seem to take the view that the relaxed state of the muscles is secondary to the prolapse, and can be improved by sphincter exercises post-operatively after the prolapsing bowel has been removed. Cohn (1942) and Gabriel (1945) have more recently sought to enhance the result of sigmoidectomy by bringing the pubo-rectales together by one or two catgut sutures inserted during the course of the operation, but it is doubtful whether a really accurate and enduring repair can be achieved by absorbable sutures in this way. Lastly, it cannot be denied that rectosigmoidectomy may lead to the formation of some peri-rectal adhesions helping to fix the rectum, but these would presumably be confined to a limited segment of bowel just above the anal canal, which would curtail their effectiveness.

ALTERNATIVE OPERATIONS AVAILABLE FOR PROLAPSE

Several of the numerous other procedures which have been described for prolapse are clearly directed against one or more of the three main aetiological factors just enumerated, and might be expected to be more effective than rectosigmoidectomy has proved to be in actual practice :

Obliteration of the Recto-vaginal or Recto-vesical Pouch by Suture or other Means.—The best known operation of this kind is that of Moschcowitz (1912) in which the pouch of Douglas is obliterated by a series of silk purse-string sutures completely surrounding the pouch and placed at intervals of 1 in. from its fundus to the brim of the pelvis. Though Moschcowitz wrote at length about this method, it is difficult to glean from his writings any idea of the late results obtaining in his cases treated by it. But Hughes (1949) succeeded in tracing 6 of 9 patients submitted to the Moschcowitz operation at St. Mark's Hospital and found that 1 had died, 4 had recurred, and 1 appeared well. Pemberton, Kiernan, and Pemberton (1953) reported on the results in 56 cases treated by a modified Moschcowitz operation at the Mayo Clinic. There was 1 operative death, and in 44 cases traced two years or longer recurrence had occurred in 5, or 11·4 per cent.

The frequent failure of this apparently rational procedure should perhaps not be entirely unexpected. For although Moschcowitz has likened the recto-vaginal or recto-vesical pouch of peritoneum to a hernial sac it would be more accurate to compare it with the sac of a *direct* inguinal hernia which is not amenable to surgical ligation and which is associated with pronounced weakness of the overlying fascia, muscles, and aponeurosis requiring surgical attention. It would be as illogical to hope for cure of a direct inguinal hernia solely by partial excision or plication of the protruding peritoneum as to expect permanent relief of a rectal prolapse by Moschcowitz's operation alone. Procedures which would seem to obviate to some extent this defect of Moschcowitz's technique are those of Mayo (1938) and Ripstein (1952) which elevate the pelvic peritoneum on a scaffolding of fascial grafts placed across the pelvic brim. But though they, like every other reported operation for prolapse, have yielded excellent immediate results in the hands of their originators, no reports of the late results in any large series of cases treated by them are available.

Excision or Obliteration of the Recto-vaginal or Recto-vesical Pouch combined with Repair of the Pubo-rectales.—This is the operation proposed by Roscoe Graham (1942) and Dunphy (1948), and it must be admitted that in theory it is a much more satisfying technique than is obliteration of the peritoneal pouch alone. In Roscoe Graham's operation both steps were carried out through the abdomen. Dunphy, on the other hand, used a perineo-abdominal method. In the perineal phase the prolapse was amputated by rectosigmoidectomy; at the same time the deep pouch of peritoneum was partly excised and the pubo-rectales muscles approximated by suture. A few days later the abdomen was opened and the remnant of the peritoneal pouch obliterated by suture as in Moschcowitz's method. Butler (1954) introduced a synchronous combined version of Dunphy's operation, the abdominal and perineal phases being performed simultaneously. He also excised redundant bowel at the same time. Brintnall (1952) and Newell (1954) have likewise favoured a combined abdomino-perineal repair for complete prolapse but without removal of any part of the rectum or colon. This latter plan has also been employed by Hughes, Gleadell, and Turner (1957).

Though it might seem that access to the muscles of the pelvic diaphragm ought to be easier from below, as in these various combined abdomino-perineal repairs, this has not been so in my experience of two operations by Dunphy's technique and of several purely perineal operations of McCann-type (1928) for repair of the pubo-rectales in cases of rectal prolapse. I have no doubt at all that suture of these muscles is much easier from above. Admittedly, the pelvic floor lies at a considerable distance from the abdominal operator and long instruments are required, but he is working through the wide end of a cone-shaped funnel and has room for manœuvre, unlike the perineal operator who, though much closer to his objective, is approaching it through the narrow end of the funnel and is thereby greatly handicapped in his manipulations. I have come to the conclusion, therefore, that the original Roscoe Graham technique is preferable to that of Dunphy on this score, and also because the avoidance of a perineal incision probably lessens the risk of wound sepsis and of consequent sinus formation which is a danger if non-absorbable sutures are employed for the pelvic floor repair.

As for the results of these operations, unfortunately there is no long-term assessment of the results in Roscoe Graham's paper. Dunphy, Botsford, and Savlov (1953) were able to report the results in 12 cases treated by Dunphy's method up to that date with no recurrence and good functional results. Hughes, Gleadell, and Turner (1957) reported

30 cases treated by a synchronous abdomino-perineal repair, with no operative deaths; most of the cases had been treated in the previous two or three years. There had been 3 recurrences up to the time of their report.

Operations to increase the Adherence of the Rectum to its Bed.—Lockhart-Mummery's (1910) operation was designed to produce adhesions between the rectum and the sacrum by packing the presacral space with gauze introduced through a post-anal incision and retained for three weeks. Lockhart-Mummery (1934) claimed excellent results for this method, but Hughes (1949) painted a very different picture. He surveyed all 33 cases treated by this operation at St. Mark's Hospital from 1931 to 1947. There was 1 operative death; 29 of the 32 operation survivors were traced, and all were found to have recurred.

Pemberton and Stalker (1939) of the Mayo Clinic did what can best be described as an abdominal Lockhart-Mummery's operation in which the rectum was mobilized from the sacrum *from above* and then elevated and held taut by a colopexy of the lower sigmoid to the left iliac fossa. In 1953 Pemberton, Kiernan, and Pemberton reported that there had been only 6 recurrences in 41 cases treated by this method in the preceding 14 years.

Orr (1947) described a method of fixing the rectum by means of strips of fascia lata attached to the sacral promontory above, and extending longitudinally on to, various aspects of the rectum to which they were sutured below. No late results were reported.

The most recent and probably most efficient way of fixing the rectum is that suggested by Muir (1955)—namely by performing an anterior resection more or less as for a carcinoma of the upper rectum. When an abdomino-perineal excision is required for recurrence of a carcinoma after a previous anterior resection, the bowel is usually found to be firmly fixed to the sacrum at the level of the anastomosis even when the recurrence is not actually situated at this level. It occurred to Muir that anterior resection might be a useful operation in the treatment of rectal prolapse, and he has employed it on 15 occasions since 1948 with excellent immediate results, no operative deaths, and only one partial recurrence to date (Muir, 1956). However, continence has been poor in a few of the cases.

I submit that from this survey there emerge three operations which have a reasonable theoretical basis and may be superior alternatives in practice to recto-sigmoidectomy in the treatment of rectal prolapse; these are the Roscoe Graham operation (or perineo-abdominal version of it), Pemberton and Stalker's operation, and anterior resection. I have no personal experience of Pemberton and Stalker's method. Anterior resection has been used by me for rectal prolapse on three occasions only, but I am much impressed by its possibilities. The method which has appealed to me as the most rational, however, is the Roscoe Graham operation, which I have employed for most of the cases of prolapse that have come under my care in the last six years, with the exception of a few elderly patients who have been treated by the Thiersch method of encircling the anus with silver wire (Thiersch, 1891; Gabriel, 1948), an operation which I regard as purely palliative. I have now done the Roscoe Graham operation 23 times, and as I believe it is the technical difficulties, anticipated or real, which have prevented this procedure from becoming popular, it seemed to me that it might be of value to describe a reliable technique for its performance, and to examine in detail the results that may be obtained by it.

THE ROSCOE GRAHAM OPERATION

The procedure now to be described is based on the principle of repair of the pelvic floor from above, enunciated by the late Dr. Roscoe Graham of

FIG. 369.—View of the interior of the pelvis at laparotomy showing the deep pouch of Douglas and the peritoneal incision along the brim of the pelvis on either side. Note the exposure of the ureters in this incision.

Toronto, but differing from his actual technique in several respects, which, in my view, alone make the operation practicable. His approach to the separated pubo-rectales muscles was apparently made through a transverse incision in the peritoneum of the depths of the recto-vesical or recto-vaginal pouch without dividing the lateral ligaments of the rectum. His main sutures of repair were placed either in these muscles or in the fascia overlying them. It is not at all clear from his account how he disposed of the deep peritoneal pouch at the conclusion of the peritoneum.

In my experience it has been quite impossible to secure anything like adequate exposure of the pubo-rectales through a limited approach of the kind described by Graham; their proper display has demanded the fullest possible mobilization of the rectum down to the ano-rectal ring exactly as in the

abdominal dissection of a Miles abdomino-perineal excision, except of course that the superior rectal vessels are not divided. I have not been prepared to accept as satisfactory any repair of the pelvic floor that does not provide unequivocal suture of the pubo-rectales muscles themselves. Lastly, I have considered it desirable at the end of the operation

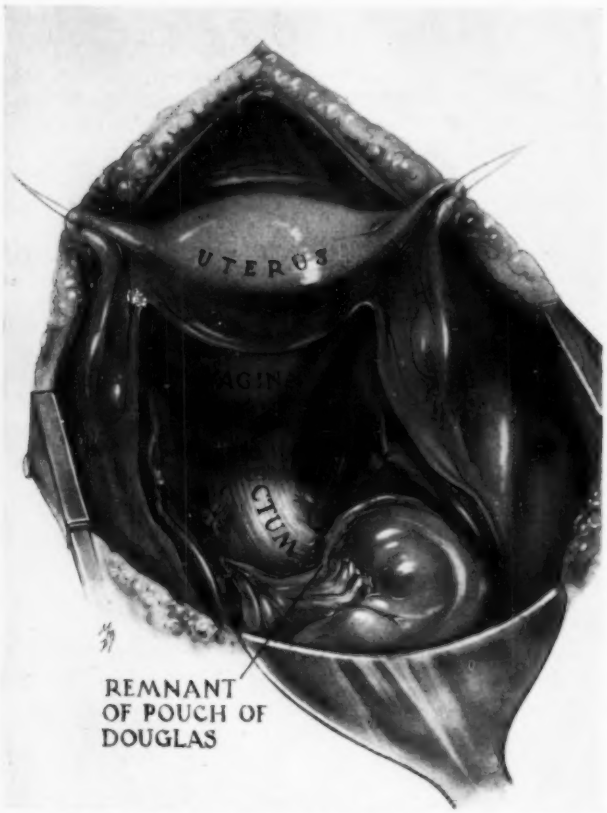

FIG. 370.—The appearance of the pelvis after the mobilization of the rectum has been completed on the anterior, posterior, and lateral aspects as far as the pelvic floor. The large pouch of Douglas has been collapsed on to the rectum and the excess of peritoneum excised up to its reflection off the front and sides of the rectum.

to arrange the pelvic peritoneum in such a manner as to eliminate the pouch of Douglas.

The operation can be performed perfectly well with the patient in an ordinary steep Trendelenburg position, but the lithotomy-Trendelenburg position, as used for a synchronous combined excision, has the advantage that it allows the surgeon or an assistant to carry out a rectal examination during the operation to verify the siting of sutures in the pelvic floor. The approach to the lower abdomen is as for any rectal excision, namely through a long left paramedian incision extending from the pubis well into the epigastric region. The wound edges are retracted with a self-retaining retractor possessing a third blade to retain the packed-off small gut in the upper abdomen. The iliac colon is now held towards the right side by the assistant while the adhesions binding it and the iliac mesocolon to the posterior parietal peritoneum of the left iliac fossa are divided with scissors. This step restores the entire sigmoid loop to its embryonic median position with a free mesocolon.

Attention is next turned to the pelvis proper. It is a useful preliminary to hitch up the uterus and adnexa by two stay sutures passed under the uterine tubes and attaching them to the towelling or skin in the pubic region. The abnormally deep pouch of Douglas is now seen to fullest advantage (*Fig.* 369). The peritoneal cut for mobilization of the rectum

FIG. 371.—Definition of the pubo-rectalis sling. The rectum is drawn upwards and forwards, and strands of connective tissue proceeding from the rectal wall are divided with scissors to expose the pubo-rectales clearly.

begins on the right side over the main inferior mesenteric artery, about the level of the bifurcation of the abdominal aorta. It is particularly to be noted that as it is carried forwards it does not descend into the pouch of Douglas, but describes a smooth curve along the brim of the pelvis, eventually reaching the midline just behind the tip of the cervix uteri. The cut on the left side begins over the left common iliac artery and is carried forwards in a similar manner, skirting the pouch of Douglas to join the right-sided incision at the cervix.

The outer edges of this circumferential incision are now elevated sufficiently to expose the ureter on either side almost down to the point where it is crossed by the uterine artery. The upper end of the left ureter is also pushed firmly laterally to separate it from the inferior mesenteric vessels, the peritoneal incision on that side being prolonged upwards if necessary to enable this to be done. This is a safeguard against ureteric injury during the subsequent dissection. The inner edge of the peritoneal cut is now brushed inwards all round so that the pouch of

Douglas is collapsed on to the rectum. As this flap of peritoneum is dissected up at the proximal end of the incision on the right side the inferior mesenteric and superior rectal vessels are exposed. If the plane of dissection is now carried just deep to them it will separate these vessels from the presacral nerve. This plane of separation is followed downwards, the

FIG. 372.—Insertion of initial deep suture in pubo-rectales. First stitch being placed in the left pubo-rectalis with boomerang needle.

two terminal branches of the nerve being gently brushed off the rectum to which they cling very closely as they separate to run to either side of the pelvis. Long scissors can now be introduced between the rectum and sacrum to open up the soft presacral connective tissue sufficiently for the hand to be passed down behind the rectum as far as the tip of the coccyx. This completes the posterior mobilization.

Attention is now turned to the anterior dissection. The posterior edge of the initial cut in the peritoneum is seized by two long forceps on either side of the midline just behind the cervix uteri, and the corresponding anterior edge and cervix are drawn forwards by a wide deep retractor. A few scissor-cuts start the dissection which is then continued with gauze pushers on long forceps so as to expose the posterior vaginal wall in front and the rectum behind. Further blunt dissection carries the separation down as far as the ano-rectal ring, bleeding from small vessels on the back of the vagina being checked by diathermy coagulation.

It only remains now to divide the lateral ligaments to complete the freeing of the rectum. So far the front of the rectum and the connective tissues stretching laterally on either side of it, constituting the so-called lateral ligaments of the rectum, are still covered by the thin but very definite fascia of Dénonvilliers. A transverse cut is made in this fascia low down over the front of the rectum, and the lateral edge of the bowel clearly defined from the adjacent fat on either side by blunt dissection with

FIG. 373.—Insertion of initial deep suture in pubo-rectales. First stitch being carried through right pubo-rectalis.

the scissors. The hand is now re-introduced into the retro-rectal space in front of the sacrum, and the index or middle finger worked forwards on the right side of the rectum to reach the point already established anteriorly, thus isolating the lateral ligament above the finger. This bulky band of tissue is then doubly clamped, divided, and ligated close to the bowel. The lateral ligament on the opposite side is next isolated and severed in the same way.

The mobilization of the rectum has now been completed (*Fig.* 370). It will be seen that it differs from the dissection employed in an abdomino-perineal excision for carcinoma only in that particular care is taken to try to avoid damage to the autonomic nerves, by dissecting out the presacral nerve and its branches above, and by keeping close to the rectum and as far away from the parasympathetic nerves on the side wall of the pelvis when dividing the lateral ligaments below.

The rectum is now drawn strongly upwards and slightly forwards, and the point of junction of the bowel and the levator muscles examined posteriorly and laterally. Usually, some further dissection of the connective tissue and fascia overlying this region

is necessary with long curved scissors, before the pubo-rectalis sling can be clearly defined as it surrounds the bowel on these aspects (*Fig.* 371). The axis of traction of the rectum is then directed more backwards and the display of the muscles continued in front of the bowel on either side.

The scene is now set for the repair of the pelvic diaphragm. The sutures, which are of strong No. 15 silk or 12-lb. braided nylon, may be introduced by a small stout half-circle needle on a long needle-holder such as Mayo's, but are better placed by a small boomerang needle, which can be much more easily manipulated in the depths of the pelvis. With the rectum pushed well backwards the initial stitch is inserted first in the left pubo-rectalis muscle and then in the right one, the point of penetration of the muscles being exactly lateral to the rectum (*Figs.* 372, 373). The result of siting the stitch so far back is that when it is drawn taut it sharply constricts the rectum, and at first sight it would appear that it must be exerting injurious pressure on the bowel. Digital examination of the rectum per anum at this stage, however, almost invariably reveals that though the bowel is in fact somewhat narrowed by the suture and the stitch itself is easily palpable through the rectal wall, there is still an ample lumen which transmits three or four fingers easily. Indeed, it has more often been found desirable to introduce another suture farther back in the pubo-rectales muscles to secure a really firm constriction of the rectum. When this first stitch is finally shown by digital examination to be satisfactory it is tied. If the tails of the knot are kept long and clipped with forceps for traction, the anterior portions of the pubo-rectales muscles are brought strongly into relief as two stout rods of muscle running forwards and laterally (*Fig.* 374). This step greatly facilitates the insertion of the remaining sutures. It is only at this stage that one realizes what substantial structures these muscles are despite their apparently atrophied condition on clinical examination. Usually three or four sutures can be placed in the pubo-rectales in the interval between the initial stitch and the vagina (*Fig.* 374, inset).

FIG. 374.—Insertion of subsequent deep sutures in pubo-rectales. Tails of first stitch have been drawn taut, causing the pubo-rectales to stand out as two prominent bars of muscle. This facilitates the insertion of the remaining sutures. Inset, the complete repair.

FIG. 375.—Alternative technique of suture of pubo-rectales *behind* the rectum.

In 2 of my 23 cases it was not possible to bring the pubo-rectales into apposition without undue tension; instead, therefore, the gap between them was bridged by a darn of nylon, taking firm bites of the muscles on either side, so placed that it narrowed the opening in the pelvic diaphragm through which the rectum passed to a size just sufficient to allow two but not three fingers to be passed on rectal examination.

On one occasion, also, I have adopted an entirely different plan of suture of the pubo-rectales, proposed by Mr. Ian Todd, to whom I am much indebted for the suggestion. This consisted of tightening the muscles by suture behind the rectum. It can be argued that this probably provides a more physiological arrangement in that it ought to accentuate

the normal forward angulation of the ano-rectal junction. In the case in which I employed this method I had had difficulty in completing the separation of the rectum from the prostate down to the top of the anal canal because of unusual bleeding. Under the circumstances it would have been difficult, if not impossible, to approximate or darn the pubo-rectales in front of the rectum by suture, and this method of stitching them behind the rectum and pushing the rectum forwards against the prostate, from which it had not been separated, overcame this difficulty for me. The actual technique of suture adopted is illustrated in *Fig.* 375. The immediate result as determined by rectal examination in this case was most gratifying, the sutures being easily palpable posteriorly in the position of the normal pubo-rectalis sling, and causing a distinct narrowing of the lumen at this level.

Reconstruction of the pelvic peritoneum is the next consideration. As previously pointed out, the entire pouch of Douglas has been displaced along with the rectum. One certainly does not want to reconstitute it, and would really prefer to excise it *in toto*. Unfortunately this is not easily done because though the front, sides, and floor of the pouch, which are composed of parietal peritoneum, are easily sacrificed, the posterior wall, which is largely formed by the visceral peritoneum covering the front of the rectum, can only be removed with difficulty by separating it from the muscle coat of the rectal wall. I have once done this, but it proved a somewhat hæmorrhagic dissection and I have never repeated it. Instead, I prefer either to excise the peritoneum of the pouch up to its line of reflection off the front and sides of the rectum, leaving the tongue of visceral peritoneum which coats the front of the rectum itself, or instead to preserve the entire pouch attached to the rectum. When the cut edge of parietal peritoneum around the pelvic brim is mobilized as in *Fig.* 376 it can be sutured to the bowel all round, as in *Figs.* 377, 378, forming a horizontal pelvic peritoneal floor covering over any unexcised part of the peritoneum of the pouch of Douglas. The rectum which lies below the level of the new peritoneal floor is unusually slack and tortuous, but no harm seems to result from this state of affairs, presumably because this infraperitoneal bowel rapidly becomes fixed by perirectal adhesions.

The abdomen is closed without drainage of the pelvic cavity.

POST-OPERATIVE MANAGEMENT AND IMMEDIATE RESULTS

There have been no operative deaths in this series of 23 patients, whose ages ranged from 17 to 82 years, and despite the fact that the operation involves an extensive pelvic dissection and much deep retraction it seems to cause little shock and to be well borne, even by the elderly. In some cases one pint of blood has been transfused during or immediately after the operation, but no more vigorous resuscitation has ever been required. Parenteral fluids have generally been terminated in 36–48 hours except in 2 patients who developed some degree of paralytic ileus, necessitating continuation of the intravenous drip and gastric suction for several days before it abated.

It has been our practice to keep the patients in bed for 3 weeks after this operation, generally in a low reclining position in order to minimize the strain on the newly formed pelvic floor till it has had a chance to become consolidated with adhesions. An in-dwelling urethral catheter has been retained for 4 days, as after an excision of the rectum, and on its removal normal micturition is usually soon restored. The bowels have been left confined for 3–4 days and then opened with a purgative such as cascara,

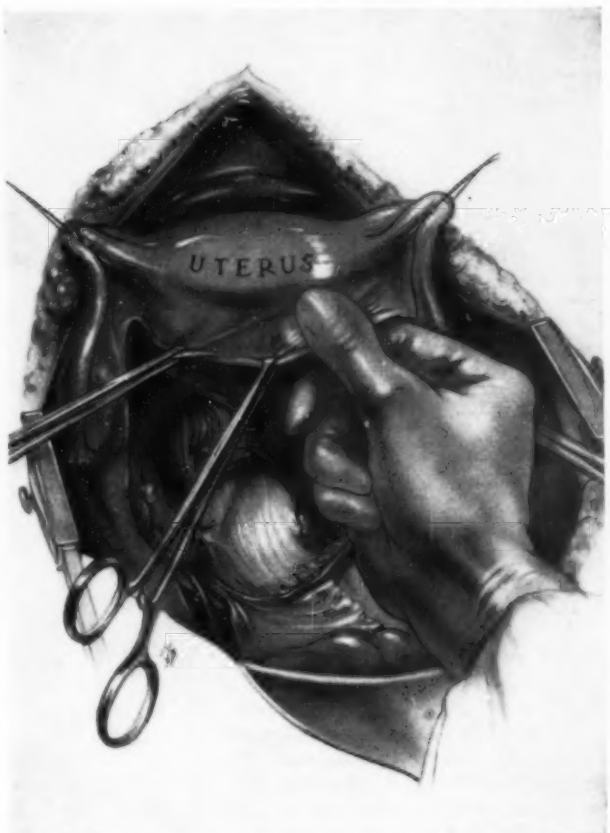

FIG. 376.—Teasing out the fringe of pelvic peritoneum preparatory to suture.

or by an enema, or glycerine suppository. This is followed by a period of irregularity often associated with some degree of incontinence. The best management of this state of affairs has been found to be a daily enema to empty the distal colon completely. By the time the patient is ready to leave hospital at the end of 4–5 weeks it has usually been possible to wean her off this routine on to natural evacuations with or without aperients, but in 5 cases it was necessary to persist with the enemata after discharge. Post-operative anal sphincter exercises have been prescribed in all cases, and the physiotherapist has attended twice daily, not only to institute regular respiratory exercises, but also to supervise these repeated voluntary contractions of the anal sphincters and in certain cases to supplement them with stimulation of these muscles with the faradic current.

Two complications require to be mentioned. In one patient there was a *deep venous thrombosis* in one leg, which soon subsided with anticoagulant therapy. Another patient developed a *pelvic abscess* which

was not recognized till the fourteenth day, when it burst into the vagina and through the lower end of the abdominal wound. This complication must presumably have been due to extraneous contamination, for the bowel is not opened at any stage during this operation. It was feared that, owing to the presence of the deep sutures of nylon in the pubo-rectales, a troublesome stitch sinus might result, but in fact the discharge soon ceased.

FIG. 377.—Start of pelvic peritoneal suture.

FOLLOW-UP STUDY

All 23 cases have been carefully re-examined at intervals up to the time of writing this report, which has meant that 2 patients have been followed for 7 years, 2 for 5 years, 4 for 3 years, 5 for 2 years, 4 for 1 year, and 6 for less than 1 year.

Objective Results.—These have been uniformly excellent, for in no case has there been any suggestion of a recurrence of the prolapse. Even with the most vigorous straining it has been found impossible for any of the patients to produce an external protrusion. Moreover, regular digital examinations of the rectum have shown that, for the first year or so in most cases and for very much longer in some, the bowel remains firmly gripped by the tightened pubo-rectales muscles and the deep sutures of repair. Whilst this remains so it would seem impossible for any prolapse to make its way past this barrier. Subsequently the constriction by the pubo-rectales often becomes less evident, but there is still no tendency to prolapse of the anterior rectal wall on straining. The actual sutures nearly always remain distinctly palpable anteriorly. They are in fact often so clearly felt that they appear to lie in the submucosa, and it is somewhat surprising that the overlying mucosa is not occasionally damaged during the passage of hard fæcal masses, with resulting ulceration, sepsis, and exposure of the sutures. However, no such late septic complication has occurred in any of these cases. But the patient already referred to, who developed a pelvic abscess in the immediate post-operative period, has had a recurrence of her discharge of pus intermittently through the lower end of her abdominal scar. This has not had any ill-effect on the cure of her prolapse, now four years after operation, but has proved a minor inconvenience to her. The persistence of the infection in this case must be attributed to the retained non-absorbable sutures in the pubo-rectales.

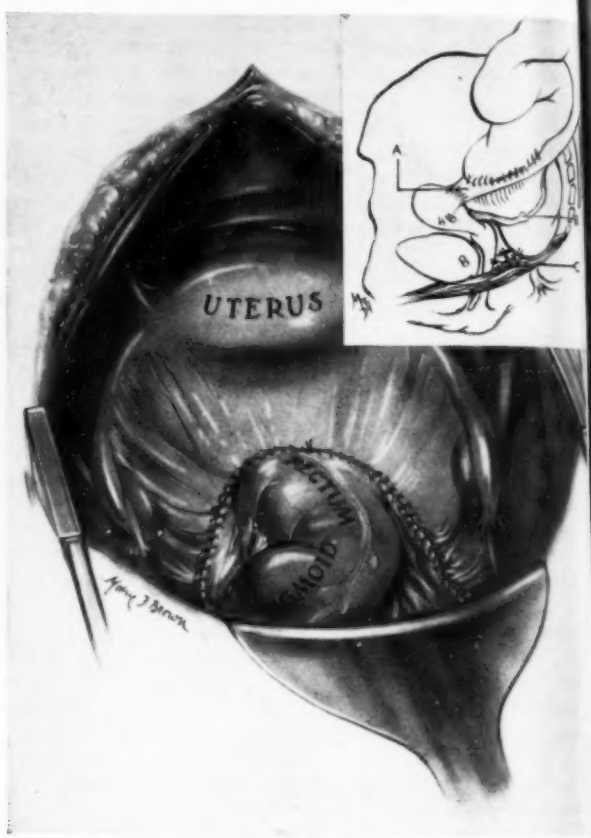

FIG. 378.—Suture of pelvic peritoneum to rectum and mesorectum completed. Inset, diagrammatic representation of relation of new pelvic peritoneal floor (A) to old remnant of pouch of Douglas (B) and to the pubo-rectalis repair (C).

The state of the anal sphincters has been much less gratifying on the follow-up. Despite the fact that most of these patients practised regular sphincter exercises for long periods after discharge from hospital, the tone and contractile power of the muscles has remained substantially unchanged. Where the state of the sphincters was good before operation it has continued so, and patients who pre-operatively had an atonic weak sphincter, capable of little or no voluntary contraction, are still in much the same condition. Of the 7 or 8 cases in this category, 4 have a patulous anus, and as 2 of

these have also some degree of internal hæmorrhoid formation, the effect is that anal mucosa is exposed at the anal orifice. In these 2 patients submucous injections of 5 c.c. of 5 per cent phenol in almond oil have been given for the internal hæmorrhoids, with some benefit.

Subjective Results.—These have been a good deal less impressive than the objective results, for several of the patients are not fully satisfied with their post-operative condition. From the point of view of function the cases can be divided into three groups: (1) Fifteen patients consider that they are completely continent under all conditions and have no complaints. (2) Four patients are usually continent, but they are troubled with frequent small motions, up to 3 or 4 per day, consisting of numerous tiny fæcal pellets. They do not usually wear a perineal pad unless they have a bout of diarrhœa, when their control is apt to break down. They are generally, but not always, continent for flatus. (3) Four patients are frankly incontinent. They usually have some fæcal soiling of the peri-anal skin on examination and always wear a pad. Two of them find that a daily enema, by forcibly emptying the colon, very much lessens their incontinence, but the others have difficulty in administering the enema owing to the patulous condition of the anus, and have not been helped by this régime.

There was, as might have been expected, a fairly close correlation between the functional result and the state of the anal sphincter apparatus in these patients. Thus most of the cases in categories (2) and (3) had grossly subnormal sphincter tone, and it is difficult to see how they could ever be continent for liquid motions. Their incontinence was, in fact, a perpetuation of a pre-operative condition, for these patients were equally incontinent before operation. This fact tended to be obscured by the presence of the prolapse itself, which, when in the descended condition, rendered anal control quite impossible. But it is clear from careful interrogation that even when the bowel had not prolapsed, many of the patients with weak atonic sphincters were quite unable pre-operatively to restrain the passage of liquid motions or flatus.

There were 2 patients who complained of being incontinent after operation despite the fact that they possessed well-developed sphincter muscles, and this suggested the possibility that they were suffering from a form of 'sensory incontinence'. No part of the rectum is resected in the Roscoe Graham operation, but the mobilization of the bowel is very extensive and it seemed not altogether unlikely that afferent nerve-fibres from the rectum, which apparently accompany the parasympathetic supply (Goligher and Hughes), might be so seriously interrupted as to impair rectal sensation. Rectal sensation was therefore treated by balloon distension, as previously described (Goligher and Hughes), as a routine post-operatively in 17 of the cases in this series, and in 12 of them also before operation. It was noted that even before operation, sensation was relatively obtuse in many of these patients, and they could often stand considerable distension of the rectum before any sensation was experienced. Post-operatively there was no decisive difference, except in one patient early in the series who seemed to be completely anæsthetic to balloon distension. As she had excellent sphincter muscles, and stated that she was not only incontinent, but unable to distinguish between fæces and flatus, this appeared to establish her incontinence as of a sensory type. But none of the other 16 patients were insensitive to balloon distension, and they claimed usually to be able to tell when the rectum contained flatus or fæces, so it can only be concluded that a sensory effect is not part of the mechanism of their poor functional result.

Urinary and Sexual Function.—There was no residual urinary dysfunction in any of these 23 patients. Of the 4 male patients in the series only one was married; he reported that following operation erections were normal or nearly so, but that there was impairment of the orgasm and failure of ejaculation, presumably due to damage to the presacral nerve. Sexual function in the other two was apparently undisturbed.

Psychological State.—It is well known that sufferers from complete prolapse of the rectum frequently exhibit psychopathic personalities, and that they are sometimes precipitated into an acute psychiatric condition by operation for repair of the prolapse. Four of the patients in this series were decidedly odd, even bizarre, individuals. There was no instance of acute mania in the immediate post-operative period, but one patient underwent a more gradual deterioration in her mental condition in the first few months following operation, and had eventually to receive institutional treatment for severe melancholia. One of the 3 other patients of rather peculiar personality seemed to improve psychologically after operative treatment of the prolapse had been carried out; the other 2 have remained unchanged.

DISCUSSION

The late results so far elicited in this series of 23 patients treated by the Roscoe Graham type of operation, though not entirely satisfactory, are certainly encouraging. Clearly a longer period of follow-up will be required on several of these patients, and preferably also on other cases as well, to determine more precisely the prospects of cure by this operation, but the fact that all 8 patients treated over three years ago still remain free from recurrence seems to establish beyond question that the good objective results of the operation can endure. This curative effect must be attributed to one or more of the three main actions of the operation—namely, the exclusion of the pouch of Douglas, the repair of the pubo-rectales, and the formation of peri-rectal adhesions—though their relative importance is a matter for discussion:—

Exclusion of the Pouch of Douglas.—We know from the work of Hughes that simple obliteration of the pouch of Douglas or recto-vesical pouch for rectal prolapse, unaccompanied by any other manœuvre, is almost invariably followed by recurrence. It might be doubted, therefore, whether the exclusion of the pouch practised in the Roscoe Graham operation can exercise much lasting benefit. On the other hand, the technique employed for eliminating the pouch of Douglas in the version of Roscoe Graham's operation just described is probably

mechanically more effective than that used by Moschcowitz. It would certainly be interesting to observe at subsequent laparotomy whether the pouch of peritoneum does ever re-form after this operation, but so far the opportunity to investigate this point has not been vouchsafed to us. At least the exclusion of the recto-uterine or recto-vesical pouch is probably of temporary value in avoiding recurrence of the prolapse till other curative factors in the operation have had a chance to become effective.

Repair of the Pubo-rectales Muscles.—In the early post-operative months, the repair of these muscles, so easily palpable on rectal examination as a firm constriction of the bowel, would seem to impose an insuperable obstacle to recurrence of the prolapse at this stage. The subsequent relaxation of the constriction in some cases, due presumably to the sutures cutting out, undoubtedly diminishes its effectiveness, but as with the excision of the peritoneal pouch, the muscle repair may have been of temporary value in restraining the prolapse until other factors have had a chance to become operative.

Formation of Peri-rectal Adhesions.—One result of the thorough mobilization of the rectum practised in this more radical version of the Roscoe Graham operation is that extensive raw areas are created, and these presumably lead to the later formation of many peri-rectal adhesions. It is highly probable that this process helps to fix the rectum to its bed and thus materially assists in the prevention of recurrence of the prolapse. It may be, indeed, that such fixation is the main mechanism of cure by the operation, and that thorough mobilization of the rectum alone, or better, combined with exclusion of the pouch of Douglas (to allow firm infraperitoneal adhesions to form between the rectum and the pelvic walls) but without suture of the pubo-rectales, would be equally curative. As the insertion of the pubo-rectales sutures is the most difficult part of the Roscoe Graham operation, it would be an attractive alteration of technique to the surgeon if this item of repair could be omitted without detriment to the result. Clearly the next step in the evaluation of the Roscoe Graham procedure would be to treat, for comparison, a number of patients with prolapse by this modified technique.

COMPARISON OF ROSCOE GRAHAM OPERATION WITH ANTERIOR RESECTION IN THE TREATMENT OF RECTAL PROLAPSE

The good results obtained by Muir in the treatment of rectal prolapse by anterior resection have already been mentioned. From his report it would appear that the results have been roughly the same as those secured by me with the modified Roscoe Graham operation, and over a similar period of follow-up. Muir, however, does not record the functional condition of his patients after operation in the same detailed fashion as employed by me, so that it is difficult to compare the functional results in the two series of cases. It may be that the choice between these two procedures will depend essentially on the respective technical difficulties associated with their performance and with the quality of the resulting rectal function. Muir records no operative mortality or significant morbidity in his cases, but it is my impression, from the three anterior resections performed by me for this condition, that a really low anterior resection for rectal prolapse is, as a rule, a more difficult operation than the same procedure for carcinoma. This difficulty is due to the fact that the lower rectum has a wide lumen and a thick wall at the point of section, not easy to control temporarily with a clamp or to turn in subsequently by suture. Without wishing to exaggerate the technical difficulties of anterior resection for prolapse I think it would be fair to say that it probably has no significant advantage on this score over the Roscoe Graham operation.

As for subsequent rectal function, it is difficult to make a valid comparison between the results of the two operations in this respect. However, one can see good theoretical reasons why the functional results might be even poorer with anterior resection than with a repair operation, for in the former a considerable segment of rectum with its important sensory function is sacrificed and, further, nothing is done directly to strengthen the deficient musculature of the pelvic diaphragm.

Lastly, it remains to consider the possible advantages of a combination of anterior resection and the Roscoe Graham repair. In so far as the results of either operation alone *qua* cure of the actual prolapse seem to be excellent, it is difficult to say what is gained by combining them. The serious disadvantage I apprehend of a combination of the two operations is that temporary leakage after low anterior resection is very common, and if this occurred in a patient who had non-absorbable sutures buried in the pubo-rectales a persistent fistula would be very likely to result.

SUMMARY

The main operations recommended for the treatment of complete prolapse of the rectum are critically examined, preference being expressed for the Roscoe Graham type of repair.

The author's technique for the performance of the operation is described in detail. It is emphasized that there are three essential steps: the thorough mobilization of the rectum; the suture of the pubo-rectales muscles; and the excision or exclusion of the abnormally deep pouch of Douglas.

Twenty-three patients have been treated by this method over the past 7 years without operative death or serious immediate complication. All have been carefully followed up, and a recent survey of the cases reveals:—

(1) That there have been no recurrences of the prolapse.

(2) That 15 patients have excellent functional results; 4 are occasionally unable to control liquid stools but can control solid motions, and 4 are completely incontinent.

It is suggested that the main curative factor in this operation is probably the formation of peri-rectal adhesions after the extensive pelvic dissection which fix the bowel to its surroundings. The elimination of the pouch of Douglas and even the repair of the pubo-rectales may help to prevent recurrence in the early post-operative stage.

The relative value of the Roscoe Graham repair and anterior resection for prolapse is discussed. It is pointed out that a combination of the two operations might be especially liable to be followed by the development of a persistent sinus from the non-absorbable sutures in the pubo-rectales.

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ACUTE CHOLECYSTITIS DUE TO GAS-PRODUCING ORGANISMS

BY W. A. WILSON, WARRINGTON

GAS-PRODUCING organisms are listed as causal factors of acute cholecystitis in most text-books of surgery and surgical pathology. None of the authors, however, describes cholecystitis due to these organisms in any detail and only one reference can be found in the British literature (Hutchinson, 1956). The condition has been reported several times in the American literature under a variety of names: emphysematous cholecystitis, pneumo-cholecystitis, pyopneumo-cholecystitis, gangrenous cholecystitis, and gas phlegm of the gall-bladder. Emphysematous cholecystitis is the term which has been most regularly used, and will be employed here.

The finding of gas in the gall-bladder at operation was first reported by Lobingier in 1908. It was Kirchmayr in 1925, however, who first gave an accurate clinical description of the condition, but it was not until after 1931 when Hegner described the radiological appearances that the condition became well known. Since then a number of cases have been reported (*Table I*). The author's own case of acute cholecystitis, treated without surgery, is reported as follows.

CASE REPORT

F. P., male, aged 43 years, was admitted to the medical wards of the Walton Hospital, Liverpool, on March 17, 1952, complaining of severe abdominal pain of 24 hours' duration, worst in the right hypochondrium. The pain had started as a "heavy feeling" in the abdomen and 12 hours later became more severe and settled in the right upper quadrant. The patient had vomited once with easing of the pain. There was nausea and some loss of appetite since the onset of the illness. There had been no previous indigestion, but some flatulence and an aversion to fats. The bowels were regular and had moved on the day of admission. There was a history of hospitalization for a similar attack 18 months previously, but this had settled in 4 days and a diagnosis of acute gastritis had then been made.

On examination, the patient looked ill, was sweating and toxic. Temperature 97·6° F.; pulse 76; respirations 20. The conjunctivæ were pale but not icteric. The tongue was furred and dry. There was no adenopathy. The breathing was shallow and caused pain in the right upper abdomen. The abdomen was tender with guarding

FIG. 379.—Shows gall-bladder outlined by gas with gas in the tissues around the gall-bladder.

Table I.—To Illustrate Points of Discussion in Reported Cases

Case Number Author and Date	Sex and Age	History of Previous Attacks	Palpable Mass	Icterus	Diabetes	Radiology	Initial Treatment	Surgery						Outcome	Cause of Death	Bacteriology
								Days after Admission	Reason for Surgery	Condition of Gall-bladder	Gas	Stones	Procedure			
1. Lobingier, 1908	M. 55	Yes	Yes	Yes	No	Not done	O.	1		Gangrenous, distended with gas	In gall-bladder	Yes, single	Cholecyst-ectomy	Recovery		
2. Tanner, 1921	M. 44	Indefinite	No	No	No	Not done	O.	1		Hæmorrhagic with patches of necrosis	In gall-bladder	No	Cholecyst-ectomy	Recovery		Streptococci + *B. coli*
3. Kirchmayr, 1925	M. 64	No	Yes	Yes	No	Not done	C.	3	Mass enlarging	Gangrenous	In and around gall-bladder	Yes, many	Cholecyst-ectomy	Recovery		*Cl. welchii*
4. Hegner, 1931	M. 62	Yes	Yes	Yes	No	Typical	C.	6	Area of gas increased	Necrotic	In and around gall-bladder	Yes, single	Cholecyst-ostomy	Death	Pulmonary embolus 4th post-operative day	Atypical *Cl. welchii*
5. Simon, 1932	M. 32	No	No	No	No	Typical	C.	6	Area of gas increased	Gangrenous and ruptured	In and around gall-bladder	Yes, many	Cholecyst-ectomy	Recovery		Sporing anaerobes
6. Wybauw, 1936	F. 53	Many	No	No	No	Gall-bladder filled with stones, outlined by gas	C.	3		Wall thickened and contained pus	In gall-bladder and in walls	Yes, many	Cholecyst-ectomy	Recovery		
7. Schmidt, 1938	M. 38	Many	No	No	No	Typical after 12 days	C.							Recovery		
8. Del Campo and Otoro, 1940	M. 74	No	Yes	No	No	Gas in common duct and biliary radicles after 5 days	C.	6	Mass enlarging + gas on X-ray	Inflamed	In gall-bladder	Yes, many	Cholecyst-ostomy	Death 4th post-operative day Calculus at ampulla on post-mortem	Generalized gas gangrene of heart, lungs, liver and kidneys	*Cl. welchii* from gall-bladder pus
9. McCorkle and Fong, 1942	M. 49	Yes	No	No	No	Gas in gall-bladder	O.	1		Gangrenous	In and around gall-bladder	Yes, single	Cholecyst-ostomy	Death after 48 hours	Fulminating gas gangrene of peritoneum and abdominal wall	*B. coli* + anaerobic bacilli resembling *Cl. welchii*
10.	M. 59	Many	No	No	No	Typical after 46 hours	C.							Recovery		*B. coli* and anaerobes of the *Clostridium* group in the duodenal bile
11.	M. 49	No	No	No	Yes	Typical 2nd and 3rd day	C.							Recovery		
12. Stevenson, 1944	M. 64	No	Yes	Yes	Yes	Typical	C.		Mass and gas increasing	Gangrenous	In and around gall-bladder	Many	Cholecyst-ostomy	Recovery		Aerobic and anaerobic streptococci
13.	M. 63	No	Yes	No	No	Typical 6th day	C.	11	Dissemination of gas	Gangrenous and ruptured	In and around gall-	Yes, single	Cholecyst-ectomy	Recovery		Aerobic cultures negative; no anaerobic

12. Stevenson, 1944	M. 64	No	Yes	Yes	Yes	Typical	C.		Mass and gas increasing	Gangrenous	In and around gall-bladder	Many	Cholecyst-ostomy	Recovery		Aerobic and anaerobic streptococci
13.	M. 63	No	Yes	No	No	Typical 6th day	C.	11	Dissemination of gas	Gangrenous and ruptured	In and around gall-bladder	Yes, single	Cholecyst-ectomy	Recovery		Aerobic cultures negative; no anaerobic culture
14.	M. 52	Many	Yes	Yes	No	Typical	C.							Recovery		
15. Hutchinson, 1946	M. 56	No	No	No	No	Not done	O.	1	Diagnosis of acute cholecystitis	Gangrenous	In gall-bladder	Two in cystic duct	Cholecyst-ectomy	Recovery		*Cl. welchii* from mucous membrane
16. Heifetz and Sentura, 1948	M. 52	Yes, one	Yes	Yes	No	Typical	C.	20	Flare-up of infection	Gangrenous	In and around gall-bladder	Many	Cholecyst-ostomy	Recovery		*Strep. fæcalis*, organism of paracoli group and bacteroides group
17.	F. 57	Many	Yes	No	No	Typical with stones	C.	12	Persistence of inflammation	Gangrenous	Gas in and around gall-bladder	Many	Cholecyst-ectomy	Recovery		*Cl. filiforme*
18. Culver and Kline, 1948	M. 60	Yes, one	No	Yes	Yes	Typical	C.							Recovery, but recurrence in 2 mth.		
19. Friedman, Aurelius, and Rigler, 1949	F. 53	Many	No	No	No	Typical	C.	11	Persistence of symptoms	Thick walled with gas and pus	In and around gall-bladder		Cholecyst-ectomy	Recovery		Staphylococci anaerobic culture not done
20.	M. 68	Yes, two	Yes	No	No	Typical after 12 days	C.							Recovery		
21.	M. 66	No	No	No	Yes	Gas in and outlining gall-bladder. Gas in ducts	C.							Recovery		
22.	F. 65	Yes, cholecyst-ostomy 38 yr. before	Yes	No	No	Gas in gall-bladder	C.							Recovery		
23. Jemerin, 1949	F. 68	No	Yes	No	No	Typical	C.	14	Improvement in general condition	Gangrenous	In gall-bladder	Yes	Cholecyst-ectomy	Death	Cerebro-vascular accident	*Cl. ædematiens*
24. Gowdey and Copeland, 1950	M. 57	Many	No	No	No	Gas and fluid level after 18 hours	C.	5	Persistence of symptoms		In gall-bladder		Cholecyst-ectomy	Recovery		
25. Retterbush, Fishman, Baird, and Collins, 1951	M. 65	No	No	No	No	Typical	O.	1		Blebs in wall and patches of necrosis	In gall-bladder	Many	Cholecyst-ectomy	Recovery		*B. coli*
26. Wilson (present case), 1957	M. 43	Yes, one	Yes	No	No	Typical	C.							Recovery		

Radiology—Typical = gall-bladder outlined by gas with fluid level in gall-bladder, and gas in surrounding tissues.
Initial treatment—O. = operative, C. = conservative.

of the right side, maximal in the right hypochondrium. There was rebound tenderness in the upper abdomen and no palpable viscera. On rectal examination there was no abnormality. The respiratory and cardiovascular systems were normal, the blood-pressure being 130/80.

A diagnosis of acute cholecystitis was made and a straight radiograph of the abdomen on the day of admission showed no abnormality. Conservative treatment was instituted with glucose fluids by mouth, pethidine, 100 mg. p.r.n., and crystalline penicillin, 250,000 units 6-hourly. His condition had not improved by the following day, when his temperature was 101° F. and his pulse 100. He was still sweating and toxic, and the physical signs in the abdomen were as before. A diagnosis of acute cholecystitis was reaffirmed, and since the inflammation was well localized and the history was of 48-hours' duration, conservative treatment was continued, and oral fluids were stopped. A Ryle's tube was inserted into the stomach, and nutrition and electrolyte balance maintained by intravenous drip infusion.

By the following day, March 19, his general condition had improved and there was less tenderness in the right hypochondrium. On March 20 a mass became palpable in the right hypochondrium, and although his general condition continued to improve, this mass seemed to enlarge for the next four days; thereafter it slowly subsided. However, radiographs of the abdomen on March 22 showed a large gall-bladder outlined by and containing gas, and in the film in the upright position there was a fluid level. The tissues round the gall-bladder were also infiltrated with gas (*Fig.* 379).

Radiological check was kept on the abdomen, and on April 1 a small amount of loculated gas was still present in the region of the gall-bladder. By April 9, this gas shadow had almost disappeared. Subsequently the gall-bladder could not be visualized by Graham's cholecystography.

The patient was discharged on April 12, feeling very well. Elective cholecystectomy was advised, but he refused surgery, preferring to wait and see if he had any further attacks. He has since been seen at a follow-up clinic and is back at work with no further symptoms, although remaining fairly strictly on a fat-free diet. The patient has had a barium meal, and this shows no abnormality of the stomach or duodenum.

DISCUSSION

Although the number of cases of emphysematous cholecystitis is small, several factors emerge which seem worthy of discussion. While the condition closely resembles cholecystitis due to non-gas-producing organisms, there are certain features which are different.

Age.—Although in the cases reported (*Table I*) two patients were in their thirties, the majority have been in the sixth or seventh decades, with an average age of 56½, somewhat older than the more common form of acute cholecystitis.

Sex.—It is generally accepted that acute cholecystitis is more common in females than males in the proportion of 5 to 1, but in emphysematous cholecystitis 21 out of 26 have been male.

Diabetes.—It is not generally agreed that cholecystitis occurs more often in diabetics, but in the reported cases of emphysematous cholecystitis at least 4, i.e., 23 per cent, have had diabetes. There seems no obvious reason for this, because the usual organisms in acute cholecystitis thrive in the presence of a sugar-rich medium just as well as the gas-producing organisms.

Clinical Features.—Although there is little in the clinical examination to distinguish cholecystitis due to gas-producing organisms from that due to non-gas-producing organisms, there runs through the case reports a statement that the patient was more toxic and more ill than the physical signs would indicate. This was certainly so in the case reported here.

There is the usual generalized abdominal pain, mainly epigastric, later localizing in the right hypochondrium. Radiation to the back and shoulders occurs, as might be expected. The attack is usually acute in onset, as in biliary colic. Nausea is almost invariable. Jaundice is not especially common, and if present is usually mild. Previous attacks of biliary colic or acute cholecystitis are common. A mass is frequently palpable. The condition takes longer to settle than the more usual form of cholecystitis and the physical signs take longer to disappear.

Radiology.—Hegner (1931) was the first to describe the radiological findings in this condition. The typical appearances are well illustrated in *Fig.* 379—an area of gas in the gall-bladder region outlining the gall-bladder (due to gas in the submucous tissues) and filling the organ, together with gas in the surrounding tissues. There is usually a fluid level in the gall-bladder as well. The diagnosis can only be made by radiology, provided that other sources of gas in the biliary system have been ruled out, i.e.:—

1. An entero-biliary fistula, usually due to a gall-stone ulcerating from the gall-bladder or common bile-duct into the bowel. A barium meal will usually clear up this doubt, as the fistula, when present, is most often between the gall-bladder and duodenum. In this condition the picture would not be complete, as there would be no gas in the pericholecystic tissue unless there was a perforation into the general peritoneal cavity as well, an event which must be exceptionally rare.

2. Reflux of duodenal gas through an incompetent sphincter of Oddi. In this case there is unlikely to be the associated clinical picture. There will be no gas in the pericholecystic tissues but there will be gas in the common bile-ducts and hepatic ducts which is not typical of emphysematous cholecystitis, although Del Campo and Otoro (1940) and Friedman, Aurelius, and Rigler (1949) did report such a finding. In Del Campo and Otoro's case a calculus was found at the ampulla of Vater, but no blockage of the common duct was reported by Friedman and others.

3. As a result of previous cholecyst-enterostomy or cholecyst-gastrostomy.

If these causes can be ruled out, the finding of gas in the gall-bladder in a patient with a clinical cholecystitis should indicate that the disease is caused by a gas-producing organism.

Aetiology.—Where, in the reported cases, the organisms have been isolated, it has usually been one of the clostridial group, and this has almost always been *Clostridium welchii* (*see Table I*).

Gordon-Taylor and Whitby (1932), in a routine examination of gall-bladders removed at operation, found *B. welchii* in 18 per cent of 50 specimens.

It is generally accepted that although the source of the organism in all cases of cholecystitis may be any septic focus, most of the bacteria come from the

intestinal tract. It is generally held that these organisms are lodged in the gall-bladder wall.

The sequence of events would seem to be—as in the usual cholecystitis—blockage of the cystic duct, usually due to stone but occasionally from inspissated bile, adhesions, or surrounding inflammation, leading to increased tension in the gall-bladder with interference with the blood-supply to, and multiplication of bacteria in, the gall-bladder wall. Areas of slough are produced in the gall-bladder if the blood-supply is sufficiently interfered with and the anaerobic bacteria flourish in the necrotic tissue.

It is suggested, therefore, that emphysematous cholecystitis is a late or secondary phase of the more usual cholecystitis, and only occurs when areas of the gall-bladder wall are necrotic and then only in the small proportion who harbour the specific organisms. This would explain the delay in the appearance of gas in a number of the reported cases.

Treatment.—Of the cases reviewed in the table, 21 of the 26 were treated conservatively initially, but operation was carried out in the course of the acute illness in 12 of these. The reasons for the change in policy were persistence of symptoms, increase in the size of the mass, or increase in the amount of gas. In one case (*Case* 23) it was because of improvement in the patient's general condition.

In those cases treated conservatively, polyvalent anti-gas-gangrene serum was used in one (*Case* 14) with recovery. This case was also treated with roentgen therapy and sulphathiazole.

Penicillin and sulpha drugs were also used in some cases, but the course of the illness in the cases where they were used did not indicate that they were of any benefit.

The main concern of treatment is to decide whether conservative or operative treatment is indicated. The number of cases under review is too small to be dogmatic on this point.

In favour of early surgery in acute cholecystitis, it may be said that it avoids gangrene and perforation of the gall-bladder, there is less subsequent morbidity, and it is usually an easy operation technically. It would seem, however, that in emphysematous cholecystitis gangrene of the wall has already taken place and, from the presence of gas in the surrounding tissues, that localized perforation has also occurred. Provided, therefore, that the localization is good, conservative treatment is to be preferred. In most of the cases reported a fairly long interval elapsed between the onset of symptoms and admission to hospital and this would militate against surgery, as it is agreed that if surgery is to be used in acute cholecystitis, it should be carried out in the first forty-eight hours.

Of the 26 cases reviewed, 5 were submitted to operation as the initial treatment, and of these one (*Case* 9) died from fulminating gas-gangrene of the peritoneum and abdominal wall. Three deaths occurred in the 12 cases which were initially treated conservatively and were later operated upon, one (*Case* 4) from pulmonary embolus, one (*Case* 8) from generalized gas-gangrene, and one (*Case* 23) from a cerebrovascular accident after a *B. coli* septicæmia. It should be noted, however, that in *Cases* 4 and 8 operation was undertaken because the mass was still enlarging, 11 and 14 days respectively after the onset of the illness, and that at the time when these cases occurred, chemotherapy and antibiotics were not available.

Of the 9 cases treated conservatively throughout their illness, there were no deaths.

It would seem that when emphysematous cholecystitis is established, conservative treatment with modern antibiotic and chemotherapeutic agents should be generally employed, but each case requires individual judgement.

The disease does not seem to be as fatal as one would expect from the virulence of the infecting agent.

SUMMARY

A case of cholecystitis due to gas-producing organisms is reported.

The literature is reviewed and some observations made therefrom, regarding clinical features, radiology, and aetiology.

The treatment is briefly discussed.

I am indebted to Dr. Robert Kemp, Consultant Physician, Walton Hospital, Liverpool, for asking me to see the case reported here.

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ACUTE TORSION OF THE GALL-BLADDER

POST-MORTEM FINDINGS IN TWO CASES

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TORSION of the gall-bladder was first described sixty years ago, and since then the number of recorded cases has amounted to about 200. In the present paper many of the case reports of the last twenty years have been analysed and 2 further instances are presented. These I believe to be the first reports in the English literature of the condition found on post-mortem examination.

FIG. 380.—The gall-bladder and liver from *Case* 2.

Historical Survey.—In 1898 Wendel reported the case of a woman of 23 years who had been under observation for sixteen months with a known wandering abdominal tumour. She had mobile kidneys, too. Following a torsion of the gall-bladder with gangrene of the wall, a local abscess formed. The condition was treated by cholecystectomy and drainage of the abscess. Only 1 recent case has resembled this original one in going on to abscess formation (Kazmann and Guthorn, 1953). By 1934 Short and Paul presented a review of 50 cases which comprised a majority of cases available in the world literature up to that time. Since then the paper by Leger, Debeyre, and Mazingarbe (1945) has been the outstanding contribution. They described 3 fresh cases and collected 91 from the literature. Husband and Schmitt (1945) presented a case and included a bibliography of 62 references, of which 25 were in English. Christensen (1954) includes a useful bibliography. Marks, Shedd, and Locke (1954) present a case with a good review of the literature. Special aspects of the condition are dealt with by Rawson (1953), by Cuervo and Cuervo (1939), who describe the disease in children, by Recht (1952), who reported torsion in a double gall-bladder, and by Marks and others (1954), who describe coincident myocardial infarction. Gross (1936), in a paper dealing with congenital anomalies of the gall-bladder, includes a section on torsion.

CASE REPORTS

Case 1.—A man aged 86 years was admitted to the West London Hospital on Nov. 28, 1956, complaining of a productive cough for a month and right-sided upper abdominal pain for a week. The upper abdominal pain was of an intermittent nature, occurring at half-hourly intervals, and it had become steadily worse. His bowels had not been open for three days, unusual for him, but he had passed flatus. For three days he had vomited after every meal. His appetite was good and he had never had digestive disturbance in the past.

ON EXAMINATION.—He was found to be a sallow old man, coughing ineffectually to bring up sputum. The tongue had a dirty yellow fur, the conjunctivæ were red. The pulse was of good volume. The abdomen was distended below the umbilicus and was tympanitic, with tenderness and guarding on the right. The liver edge was palpable, but the area of dullness was not increased. Rectal examination revealed no tenderness, only faecal lumps. Hernial orifices were normal. A diagnosis of large-bowel obstruction was made, possibly due to constipation or to a neoplasm with surrounding inflammation. His condition deteriorated suddenly and after showing the clinical picture of coronary thrombosis he died shortly after admission to hospital.

AT AUTOPSY.—Post-mortem examination was carried out next day by Dr. Pearce. The principal anatomical findings were as follows :—

The body of a thin elderly man. Cerebral softening was present. There was marked atheroma of the coronary arteries with extreme stenosis at many points and a thrombus was found in the left circumflex artery associated with a subintimal hæmorrhage. In the abdomen a little yellow fluid was present and the serosa of the intestine was injected. The gall-bladder was intensely congested and distended following torsion.

No histological report is available. The cause of death was given as : (1) Coronary thrombosis due to atheroma ; and (2) torsion of the gall-bladder.

Comment.—This case resembles that of Marks and others (1954) in which the dual pathology of coronary thrombosis and torsion of the gall-bladder coexisted. Although the myocardial circulation is influenced reflexly by distension of the biliary apparatus (see recent experimental work by Cullen and Reese, 1952), these two cases appear from the literature to be exceptional and may be attributed to coincidence.

Case 2.—A man of 81 years was admitted to Farnborough Hospital on Jan. 6, 1956, having fallen from a chair two days previously. Radiography showed a pretrochanteric fracture of the right femur and gross osteo-arthritis. He became weaker, developed bronchopneumonia and heart failure, and died suddenly on April 30, 1956.

AT AUTOPSY.—Post-mortem examination was carried out next day by myself, and *Fig.* 380 shows the fresh post-mortem appearance of the gall-bladder. The relevant findings were as follows :—

The heart was hypertrophied and the coronary arteries were atheromatous though not occluded. A little blood-stained fluid was found in the right upper abdominal compartment, where there was early fibrinous

peritonitis of the intestine in contact with the gall-bladder. The liver was small, tawny, and finely nodular. The gall-bladder was a large red organ about four times the normal size, which had undergone a complete torsion of its pedicle (*Fig.* 380). It contained altered blood but no stone. The main bile-duct was patent and contained blood-stained bile.

Histological examination showed cirrhosis of the liver and hæmorrhagic necrosis of the gall-bladder.

DISCUSSION

Most of the recent cases published in the literature have been summarized in *Table I* and include the 2 cases here recorded.

This series shows once again that it is a disease of old people (*Fig.* 381), commoner in women.

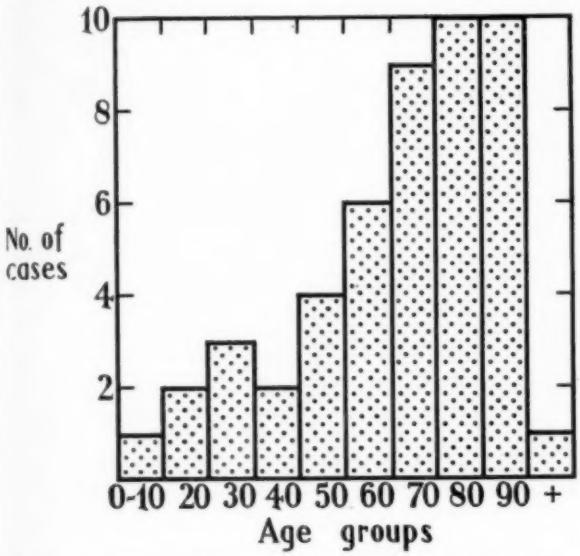

FIG. 381.—The age incidence of torsion of the gall-bladder in 48 cases.

Many of the patients are thin and visceroptotic. The onset is usually sudden and a preceding history suggestive of biliary tract disease is exceptional. The presentation of the disease is that of an acute abdominal emergency, with or without shock and the development of right-sided peritonitis and perhaps a mass. Christensen (1954), analysing the pre-operative diagnosis in 54 cases, produced the following chart :—

PRE-OPERATIVE DIAGNOSIS IN 54 CASES

Cholecystitis, mucocœle of the gall-bladder, or empyema	17
Intestinal obstruction	5
Appendicitis	20
Cancer of colon or stomach	3
Perforated ulcer	6
Twisted ovarian cyst	2
Intermittent hydronephrosis	1

A similar analysis is given by Short and Paul (1934) and no useful purpose is served by so studying the present series.

Short and Paul were quite optimistic about the feasibility of the pre-operative diagnosis of the disease : "The acute pain and vomiting, without jaundice, in an elderly female and the appearance *within a few hours of the onset* of the greatly enlarged and palpable gall-bladder, are highly suggestive. If as in one of our cases and in a few others *the tumour comes and goes* it can scarcely be anything else." But it is the exception to know the time of appearance of a mass and in only about one-third of the cases was a mass detectable. In only 1 case did the swelling come and go. And here it is of interest to record that in 3 of the 19 cases a mass was felt only under anæsthesia. (It is not recorded in those in which no mass was felt whether an examination under anæsthesia was carried out.) Krabbel (1920) made the diagnosis once, because he had seen an almost identical case a week before, but a year later he missed the diagnosis in a third case.

Is it diagnosable? The symptomatology and physical findings in the 'near miss' are those of : (1) the acute abdominal emergency ; and (2) acute inflammation of the gall-bladder. As with other abdominal emergencies in old people, up to a week or more elapsed in some cases before operation was performed, yet perforation was extremely rare. This is similar to the finding in acute cholecystitis. The non-operative management of acute cholecystitis is usually adequate because of the resistance to perforation of the distended inflamed gall-bladder.

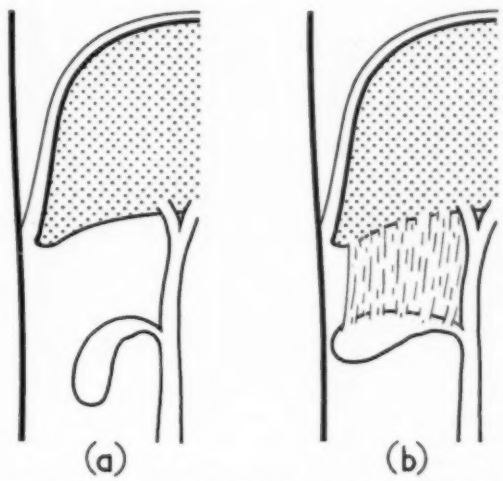

FIG. 382.—The anatomical varieties of gall-bladder that can undergo torsion : (a), pedunculated, and (b), on a mesentery.

Table I.—ANALYSIS OF 48 CASES OF TORSION OF THE GALL-BLADDER SHOWING SEX CHARACTERISTICS, WHERE RELEVANT

		Male	*Female*
Total number of cases	48		
Sex incidence		16	32
Average age		54·5	66
Mass felt	19		
Gall-stones present	16	0	16
Number of post-operative deaths	5		

The mortality of the disease according to Arthur (1937) is about 16 per cent, but it is now very much less. Four of the five deaths in the present series occurred in special circumstances :—

With coincident large myocardial infarct (Marks and others, 1954) ;

With gangrene of the gall-bladder after a seven-week history (Moses and Hicks, 1948) ;

With coexisting biliary calculus obstruction of the ampulla (Leger and others, 1945) ;

With embolic gangrene of the leg on the second post-operative day (Husband and Schmitt, 1945).

In the uncomplicated case it should be less than 5 per cent. All authors comment on the ease of performing the cholecystectomy and the smoothness of the convalescence.

AETIOLOGY

The aetiology of the disease is only partly known. Only gall-bladders which possess a mesentery or which hang free with the mesentery restricted to the cystic duct can undergo torsion (*Fig.* 382). The former is said to occur in 5 per cent of individuals (Gross, 1936); the latter is rare, but the incidence is not recorded. I think that a mesentery is sometimes acquired as a result of liver atrophy—e.g., the brown atrophy of ageing or of cirrhosis as in my second case, with consequent redundancy of the capsule. It would be interesting to record the type of gall-bladder found at different ages to see whether there is in fact an increased incidence of gall-bladders with a mesentery with advancing years.

Assuming the presence of these varieties of gall-bladder, why do they undergo torsion? The explanations found in the literature that peristalsis in the colon, stomach, or duodenum when sufficiently vigorous would initiate a twist in the pedunculated or suspended organ cannot be investigated; but there is no reason to think that the gall-bladder in these cases is in a more vulnerable condition than that part of the intestine which has a mesentery and which is in no way predisposed to volvulus. Other bodily structures which undergo spontaneous torsion are solid, e.g., the testis or subserous fibroid, or at least rendered temporarily rigid by their contents, as in the sigmoid colon. Clearly the organ itself is the twisting agent and the heavier it is the more effective. In the gall-bladder the increase in weight may be due to stone or a sudden increase in its fluid contents due to obstruction of the cystic duct.

Stones were present in half the female cases. The incidence of gall-stones as a chance post-mortem finding is much lower than this, in the region of 5 per cent. Perhaps a mild cholecystitis with cystic duct obstruction was a cause of increased weight in others. Obstruction of the venous return by torsion of the vascular pedicle is the cause of the hæmorrhagic infarction found in these cases.

SUMMARY

Two cases of torsion of the gall-bladder found at post-mortem are presented and recent literature is reviewed.

Sufficient cases are now available for study to reach the following fresh conclusions:—

1. The disease in its typical form almost eludes diagnosis.
2. The mortality is low.
3. The precise mechanism of torsion, as in other conditions, remains unexplained.

Acknowledgements.—I should like to thank Mr. C. C. Cookson, of Farnborough Hospital, and Mr. G. B. Woodd Walker, of the West London Hospital, for permission to publish details of the cases under their care; and Professor R. S. Pilcher of the Department of Surgery and Professor G. R. Cameron of the Department of Morbid Anatomy of University College Hospital Medical School for their help and criticism.

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A MODIFIED BANCROFT OPERATION FOR DUODENAL ULCER

By GEORGE QVIST

ROYAL FREE AND WILLESDEN GENERAL HOSPITALS

EXCLUSION of the pyloric antrum for duodenal ulcer was first described by von Eiselsberg in 1910. His operation consisted of simple transection of the stomach; the pyloric segment was closed and the cardiac end anastomosed to the jejunum. He described the method thus: "In cases in which gastroenterostomy is carried out on account of bleeding from a fresh ulcer of the pylorus and where I have considered that the ulcer is still not quite safely excluded, . . . I have been led to extend the gastroenterostomy to a unilateral pyloric exclusion. Before or after the gastroenterostomy, which is carried out in the typical manner, the stomach is seized between two clips on the cardiac side of the ulcer . . . and cut through, and each of the lumina sutured blind into itself."

FIG. 383.—Mobilization of stomach extended to a point 3 cm. from the pylorus to preserve arterial branches.

FIG. 384.—Crushing clamp applied transversely, and stomach divided at least 10 cm. from pylorus.

This procedure was modified by Finsterer in 1918 by the addition of sleeve resection, thus converting von Eiselsberg's "exclusion gastroenterostomy" into "exclusion gastrectomy". Finsterer stated: "The fact that in duodenal ulcer, after unilateral pyloric exclusion by von Eiselsberg's method, peptic ulcer can occur at the site of anastomosis, induced me to remove in all cases at least a half to three quarters of the stomach."

In this article Finsterer advocated excision of the antral mucosa. He wrote: "If the ulcer is so large that it extends from the papilla to the pylorus, the division of the stomach itself must be made immediately next to the pylorus; the little stump of the stomach will be closed blind, after the excision of as much mucosa as possible."

It is clear that Finsterer did not regard excision of the antral mucosa as an important step. In a later publication describing 14 years' experience of the exclusion operation (Finsterer and Cunha, 1931) there is no reference to mucosal excision except as a technical detail for adequate closure of the stump in certain cases. The operation is described as follows: "The antrum is severed two fingerbreadths centrally from the pylorus, the distal part is closed by suture in two layers and is then covered by the gastrocolic omentum. . . . the distal part, held by the clamp, is first closed with a through-and-through suture and then covered over with Lembert sutures. . . . In a case in which there is well-marked hypertrophy of the musculature the line of separation of the antrum must be further away from the pylorus (three fingerbreadths) in order to make the Lembert sutures without any stretching or tension. In these cases the mucous membrane from the eliminated portion of the antrum is removed as far as the pylorus and there closed by a suture. The seromuscularis is cut on the side of the greater and lesser curvature making thus an anterior and a posterior flap which is overlapped and fastened by suture over the mucous membrane suture in order to make a safe and sure closure."

Important contributions to the subject were made by Devine (1925, 1928) and Ogilvie (1936, 1938), but for some years gastrectomy with simple antral exclusion occupied a debatable role in the surgery of 'irremovable' duodenal ulcer.

In 1932 Bancroft described exclusion gastrectomy with 'coring-out' of the antral mucosa as a deliberate and important step in technique and established the operation on a sound physiological basis (Bancroft, 1932, 1951). Various modifications of the Bancroft operation have been described relating chiefly to closure of the seromuscular antral remnant, but on the whole antral exclusion gastrectomy has never achieved great popularity, probably because of the difficulty of ensuring really satisfactory primary healing of the antral stump. Nevertheless, the operation has a useful place in the surgery of large penetrating duodenal ulcer, the late results are good (Maingot, 1955; Makkas and Marangos, 1949), and it is felt that if sound primary closure of the antrum could be expected in every case the Bancroft type of operation would be employed much more frequently.

The fate of the seromuscular remnant of the pyloric antrum is, of course, a minor consideration from the functional aspect of successful gastrectomy,

but its efficient closure is the most important technical feature of the operation. Wider at its mouth than at its base, the gastric cuff is not well suited for closure by any standard technique and various special methods of dealing with this problem have been published by Wolfson and Rothenberg (1938), Wangensteen (1942), Allen and Welch (1942), and others. The most popular modification of the Bancroft operation is that described by Plenk in 1936; it is recommended by Maingot (1957), Tanner (1951), and Farquharson (1954).

In general, these operations consist either of inverting the seromuscular layer or of obliterating the antral space without inversion. Neither of these methods is completely satisfactory; the bulk of tissue is wrongly shaped for inverting, while obliteration without inverting stitches is contrary to the well-established Lembert principle of healing by

FIG. 385.—Seromuscular coat divided in the shape of a V, anteriorly and posteriorly.

FIG. 386.—The mucosa of the antrum ' cored-out ', including the two seromuscular wedges of tissue.

FIG. 387.—The mucosal cuff is freed down to the pylorus, clamped across with straight artery forceps, and removed.

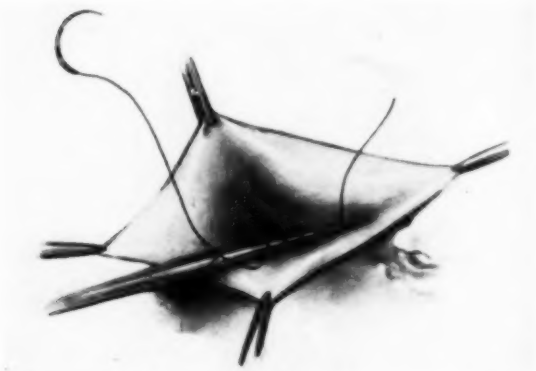
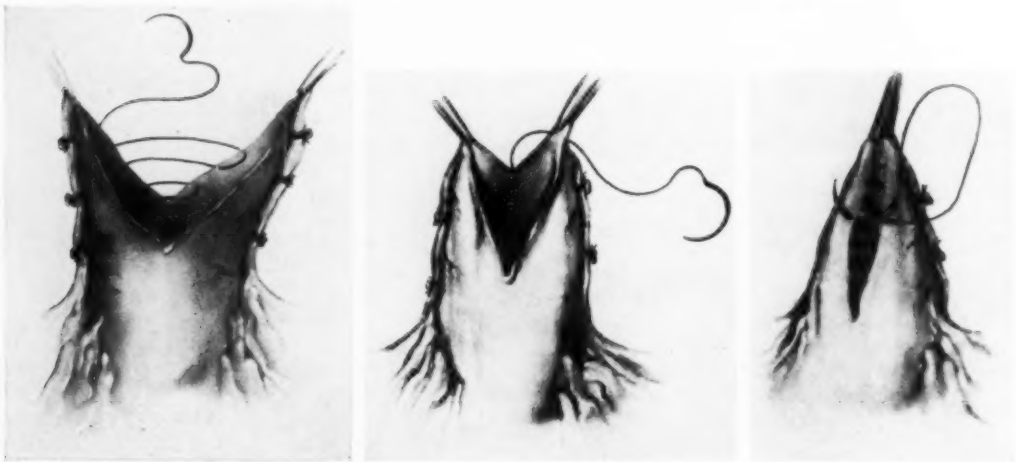


FIG. 388.—The mucosa is closed by a continuous catgut stitch in two layers.



A B C

FIG. 389.—A, The seromuscular remnant is closed by suturing the edges of the two V cuts, forming B, C, a conical bag with the apex formed by the junction of the two curvatures of the stomach.

peritoneal apposition. Post-operative hæmatoma and biliary fistula are not infrequent and indeed might be expected from these methods.

The technique here described is presented as a safe and simple method of ensuring primary closure of the antrum. It is based on three accepted essentials for adequate suture of a blind loop of bowel, viz., good blood-supply, the seromuscular inverting stitch of Lembert, and a suitable anatomical shape for inversion.

The method consists of altering the shape of the seromuscular antral remnant from a wide-mouthed bulky mass into a conical vascular stump which can be inverted easily with Lembert sutures, like an appendix or duodenal stump.

Closure of the antrum by this technique conforms to standard surgical practice and primary healing may therefore be expected in every case.

The operation has been used in 23 cases of 'irremovable' duodenal ulcer, i.e., those cases of large penetrating ulcer, often with œdema of the pancreas and stomach wall, in which standard division and closure of the duodenum was considered to be impossible or unsafe.

Primary healing has occurred in every case with no evidence of even the smallest post-operative biliary fistula.

TECHNIQUE

As has been emphasized by various authors (Malkas and Marangos; Tanner) the decision to perform antral exclusion must be taken at the beginning of the operation before there has been any interference with the blood-supply of the pyloric region.

The stomach is mobilized by division of the omenta along the greater and lesser curvatures. To the right this process is extended to a point about 3 cm. (1¼ in.) from the pylorus, in order to preserve the branches of the right gastric and gastro-epiploic arteries to this area (*Fig.* 383).

A large crushing clamp is applied transversely across the stomach about 10 cm. (4 in.) from the pylorus, and the stomach is divided above the clamp (*Fig.* 384). It is important to retain an adequate length of the pyloric segment at this stage. The mobilized antrum is atonic and overstretched, but when freed from the mucosa the seromuscular cuff retracts by about one-third in length. Unless at least 10 cm. of the atonic antrum has been retained inversion at a later stage may be difficult.

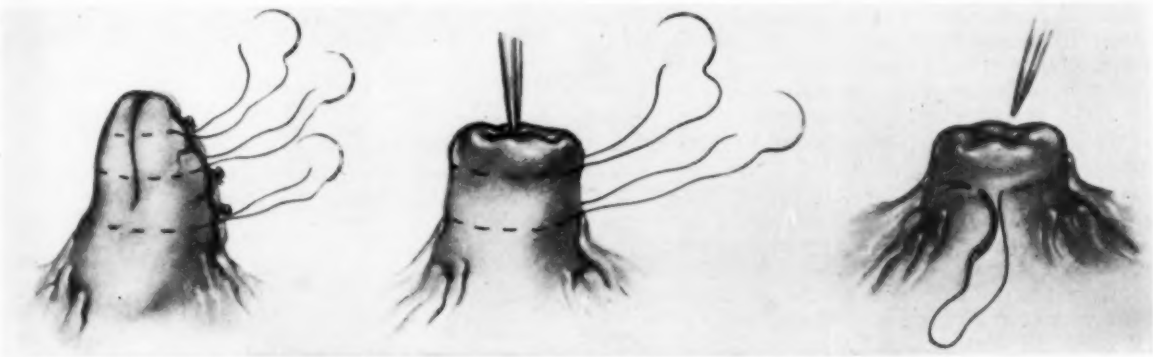


FIG. 390.—The apex of the conical stump is inverted by three seromuscular purse-string sutures.

FIG. 391.—Final stage. The retained greater and lesser omenta become easily available for oversewing the stump.

The seromuscular coat on the anterior surface of the stomach is now divided down to the submucosa in the shape of a V, the two equal limbs of the V commencing one at each curvature of the stomach and meeting at a point about 4 cm. from the clamp (*Fig.* 385). A similar C-cut is made on the posterior surface of the stomach, continuity being established at the curvatures.

The mucosa of the antrum is then 'cored-out' as described by Bancroft, the mucosal 'core' including at its free margin the two seromuscular wedges of tissue (*Fig.* 386).

The mucosal cuff is freed completely down to the pylorus where it is clamped across with a straight artery forceps and removed (*Fig.* 387). The mucosa is closed by a continuous catgut stitch in two layers in the standard way (*Fig.* 388). The amount of gastric mucosa left is therefore only that included in the artery forceps and this tissue has been crushed.

The seromuscular remnant is now closed by suturing together the edges of each of the two V cuts. In this way a conical bag is formed; it is about 7 cm. long, a good blood-supply enters its base, and the apex is formed by the junction of the two curvatures of the stomach (*Fig.* 389).

The apex of the conical stump is inverted by three seromuscular purse-string sutures (*Fig.* 390). The third of these should be inserted not less than about 3 cm. from the pylorus because beyond this point the antral stump is not 'invertible', being tethered at its base to the pyloric ring. If insufficient length of antrum has been retained and the conical stump is less than 6 cm. long, it may be difficult to insert the third suture without undue tension, and it should be supplemented or replaced by a series of interrupted sutures.

At this stage the retained greater and lesser omenta become easily available for oversewing the stump (*Fig.* 391).

It is advisable to use a stab drain in the right hypochondrium for a few days.

The type of suture material seems to be unimportant; interrupted and continuous catgut, silk, and linen thread have been used in different cases without any noticeable difference in results.

SUMMARY

A simple technique to ensure sound primary healing of the antrum in Bancroft's operation is described.

The operation consists of seromuscular inversion of a conical stump possessing good blood-supply.

Closure of the antrum by this technique is based on standard surgical principles and primary healing may therefore be expected in every case.

The method has been used without mortality or biliary fistula or any other complication in 23 cases and is recommended with confidence for the treatment of 'irremovable' duodenal ulcer.

I am grateful to my medical colleagues, Sir Daniel Davies and Dr. Frances Gardner at the Royal Free Hospital, and Dr. T. Pearse Williams and Dr. A. Wingfield at the Willesden General Hospital, for their co-operation in the management of several of these cases.

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THE EXTRAVESICAL ECTOPIC URETER

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THE ureter which drains into some site other than the urinary bladder is a relatively rare anomaly, only 459 cases having been reported at the time of writing. Although well recognized by urologists, the delay in diagnosis of many cases reported in the literature suggests that the condition is not one with which the medical profession is generally familiar. This is not surprising since any one person not dealing primarily with urological cases is unlikely to see more than one in a lifetime.

Although the condition may be symptomless in males, the incontinence usually present in the female is a great handicap to normal social activities. If labelled 'neurotic' and referred to a psychiatrist, as is not uncommon, the unfortunate sufferer is condemned unjustifiably to an abnormal existence when rapid cure could be achieved. This paper is a review of the literature up to September, 1956, and presents 3 further cases seen by the author together with 3 additional cases hitherto unreported. A possible explanation of some of the anomalies based on embryology is also put forward.

EMBRYOLOGY

The normal upper urinary tract develops as an outgrowth, the ureteric bud, from the mesonephric duct near its junction with the cloaca. This bud continues to grow in a cranial direction and at its upper end comes into contact with the metanephrogenic cap which is to form ultimately the secretory portion of the adult kidney. The ureteric bud divides at its upper end into two portions, each of which subdivides repeatedly until thirteen or more generations of collecting tubules are formed. Each of these individual collecting tubules has its own portion of the metanephrogenic blastema. Subsequent fusion of the elements of the metanephrogenic blastema and absorption of many generations of collecting tubules results in the formation of the adult kidney, calices, and pelvis.

At the caudal end of the ureteric bud the changes are more complex. The mesonephric duct below the origin of the ureteric bud becomes incorporated in the wall of the primitive urogenital sinus, its subsequent fate varying in the two sexes. It is now generally agreed that the trigone of the bladder, the supramontanal portion of the prostatic urethra, and the posterior wall of the female urethra are of mesonephric origin and not cloacal remnants as suggested by Chwalla (1928). Absorption of the mesonephric duct into the developing bladder wall together with maximal growth activity at the upper and outer lip of the mesonephric duct orifice results in lateral displacement of the ureteric orifice and an increase in the distance between it and the orifice of the mesonephric duct (Brockis, 1952).

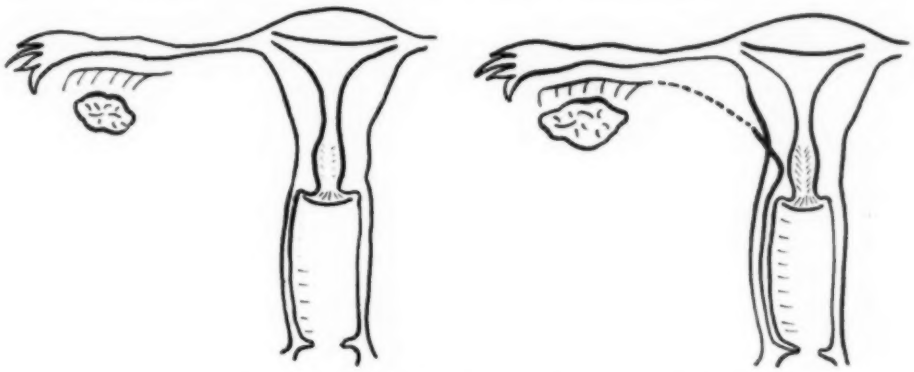

FIG. 392.—Mesonephric duct remnants in the female.

Abnormalities of development can occur at either end of the ureteric bud in association with each other or separately. Failure of complete fusion of the metanephrogenic blastema results in the persistence of fœtal lobulation or in varying degrees of sequestration of the kidney. Incomplete cranial growth of the ureteric bud leads to a low-lying pelvic or sacral kidney. Developmental arrest of the mesonephric duct between the fifth and seventh week of intrauterine life has been shown experimentally to lead to rapid regression of the metanephrogenic cap, thus causing hypoplasia or even aplasia of the kidney, while such conditions as horseshoe kidney and crossed ectopia can result from imperfect development of the metanephrogenic cap from the urogenital ridge.

At the caudal end of the ureteric bud early division and incorporation into the bladder wall will lead to the presence of multiple openings, an anomaly which will also occur if two or more buds develop from the mesonephric duct.

In the case of multiple orifices, the first to be formed lies above and lateral to any others since these are incorporated into the bladder wall later and therefore lie in the line taken by the migrating mesonephric duct orifice.

These secondary orifices in practice are nearly always associated with the upper pole of the kidney, which drains therefore into the inferior ureteric orifice. Growth disorders occurring in this region at various stages may result in a ureteric orifice which opens below the level of the internal sphincter of the bladder into some part of the mesonephric duct or its derivatives. The associated development of the paramesonephric ducts, particularly in the female, which cannot take place in the absence of the mesonephric ducts (Grünwald, 1941), will be mentioned later.

SITES OF ECTOPIC ORIFICES

In the presence of multiple congenital defects, ureteric orifices have been reported opening on the anterior abdominal wall or into cloaca, rectum, and Fallopian tubes. In these cases the defects are incompatible with life and will not be discussed.

The majority of cases drain into some part of the genito-urinary tract. In the male the commonest site is the posterior urethra above the level of the verumontanum, less commonly into the seminal vesicle, vas deferens, or ejaculatory duct. All these organs are of mesonephric duct origin. A few cases are on record where the opening was found in the prostatic utricle which is derived from the paramesonephric duct. To account for this it is postulated that a communication occurs between these two ducts as they develop in their combined tissue sheath.

In the female the ectopic orifice is usually found in the posterior wall of the urethra, vestibule, or vagina. Other reported sites are the cervix, uterine cavity, a urethral diverticulum, or Gärtner's duct.

The paramesonephric ducts in the female are the precursors of the Fallopian tubes, uterus, cervix, and the vagina as far as, and including, the upper layer of the hymen; the posterior wall of the urethra and Gärtner's ducts are of mesonephric origin, whereas the vestibule and vagina below the level of the hymen, and including its lower surface, are developed from the endoderm of the urogenital sinus. It is of interest here to note the normal fate of the mesonephric duct in the female. In approximately 75 per cent of cases the epoöphoron which lies between the two layers of the broad ligament in its lateral portions is the only recognizable remnant. In the remaining 25 per cent its extent varies. If it persists in its full length the duct of the epoöphoron passes downwards and medially towards the internal uterine os, piercing the muscle wall obliquely and approaching the lining mucosa. It then passes to the supravaginal portion of the cervix and descends in the lateral or anterolateral vaginal wall to the level of the

hymen, where it opens at its free margin. The supravaginal and vaginal portions are commonly present and form Gärtner's duct. Microscopical examination of this region has shown that the duct is often dilated and that the lining may even be fused with that of the cervical canal. The mucosa lining the duct is not constant since cubical, columnar, squamous, and transitional epithelium have all been described (*Fig.* 392).

On the basis of these findings it seems to be likely that ectopic orifices opening into the cervix, cervical canal, or vagina could best be explained by a breakdown of this thin wall of the mesonephric duct into these sites. It is common on cystoscopy to note an absence of the hemitrigone on the side where a solitary ectopic ureter is present. This suggests that the mesonephric duct with its ureteric bud has not been incorporated into the bladder and that what appears to be a ureter with an ectopic opening is in fact a complete ureteric bud together with the caudal portion of the mesonephric duct which has persisted in whole or in part. Proof of this hypothesis would seem to be difficult. Conversely it could be disproved by finding evidence of a mesonephric duct in addition to but separate from the ectopic ureter.

The relative frequency of the sites of the ectopic orifices is shown in *Table I*.

Table I.—Distribution of 494 Ectopic Ureters (including Autopsies)

Males, 128	
Posterior urethra	60
Prostatic utricle	13
Seminal vesicle	42
Ejaculatory duct	7
Vas deferens	6
Females, 366	
Urethra	129
Vestibule	124
Vagina	90
Cervix or uterine cavity	18
Gärtner's duct	3
Urethral diverticulum	2

AGE AT DIAGNOSIS

Of the cases reported in the literature, many have only been discovered at post-mortem examination. *Table II*, however, shows that in both sexes there is wide variation in the duration of the condition before it is finally recognized in life. This is all the more remarkable in the female where lifelong incontinence is usually the presenting symptom.

ASSOCIATED UPPER URINARY TRACT ABNORMALITIES

Unfortunately full details of the upper urinary tract have not been recorded in all cases reviewed in the literature, so that accurate figures cannot be obtained. The variations encountered are so numerous and have such an important bearing on treatment that wherever possible an accurate knowledge of the anatomy should be obtained prior to operation.

Although the renal element drained by the ectopic ureter has up to the present time always been on the same side as the orifice, this latter may be so close to the midline that it is impossible to be certain on which side it lies without defining the associated ureter.

Some of these variations are reproduced diagrammatically in *Fig.* 393. To these should be added cases associated with hypoplasia of the associated kidney and a unique case of crossed renal ectopia with an ectopic ureter draining the upper pole of the renal element on the correct side (Pia, 1952). Blind ectopic ureters without any evidence of associated renal tissue have also been recorded.

By far the commonest renal abnormality is that in which the upper pole of the kidney drains by a separate pelvi-ureteral system to the ectopic orifice. This renal element may be fused to the remainder of the kidney or loosely attached to it by a thin layer of fibrous tissue allowing easy separation at operation.

Table II.—Extravesical Ectopic Ureter: Age at Diagnosis in 200 Cases

Age	*Male*	*Female*	*Total*	*Percentage*
0–10	9	61	70	35
11–20	7	44	51	25·5
21–30	8	39	47	23·5
31–40	4	11	15	7·5
41–50	7	4	11	5·5
51–60	1	1	2	1
61–70	1	2	3	1·5
71–80	1	0	1	0·5
Total	38	162	200	100

Its blood-supply is also commonly separate, being derived either from the abdominal aorta just above the main renal artery or from the latter itself. This branch is the apical segment artery as described by Graves (1954).

SYMPTOMATOLOGY

The essential anatomical difference between the ectopic orifice in males and females lies in its relation to the external vesical sphincter. In the male it is always proximal to it, whereas in the female it lies distal. For this reason it is simpler to consider the condition separately in the two sexes.

In the male incontinence of urine is not a feature, although a constant trickle of urine into the posterior urethra may cause a frequent desire to micturate, with the passage of only small quantities of urine. In the majority of cases diagnosed, advice has usually been sought because complications have supervened. Recurrent urinary tract infections, prostatitis, and calculi are discovered and only after full and careful urological examination (often repeated) is the causative anomaly recognized.

If the ureter drains into a seminal vesicle rectal examination may show this to be distended and subsequent injection with contrast medium will often reveal the true nature of the condition.

Although the reported incidence in males is only 26 per cent, the greater difficulty in diagnosis makes it likely that this figure is inaccurate and should be higher. Of the 42 cases with openings into the seminal vesicles, 32 have been discovered only at autopsy.

In the female, diagnosis is relatively easy if the classical history can be obtained, but accurate detail of the anatomical abnormality may be difficult to define for reasons to be considered later.

The classical history is of a patient who has been incontinent from birth but also has normal micturition and is conscious of bladder distension. In some cases it is noticed that the incontinence is worse by day than by night. It has been suggested that this feature is due to dilatation of the ectopic ureter, which therefore acts as a reservoir during recumbency. This phenomenon, however, has also been observed in the absence of any upper urinary tract dilatation.

of infection, owing to repeated straining efforts to empty the bladder.

COMPLICATIONS

It is well known that congenital abnormalities of the urinary tract are more prone to disease than the normal. In these cases obstruction, infection, and calculus formation are very liable to occur. The ectopic orifice is usually smaller than normal, the

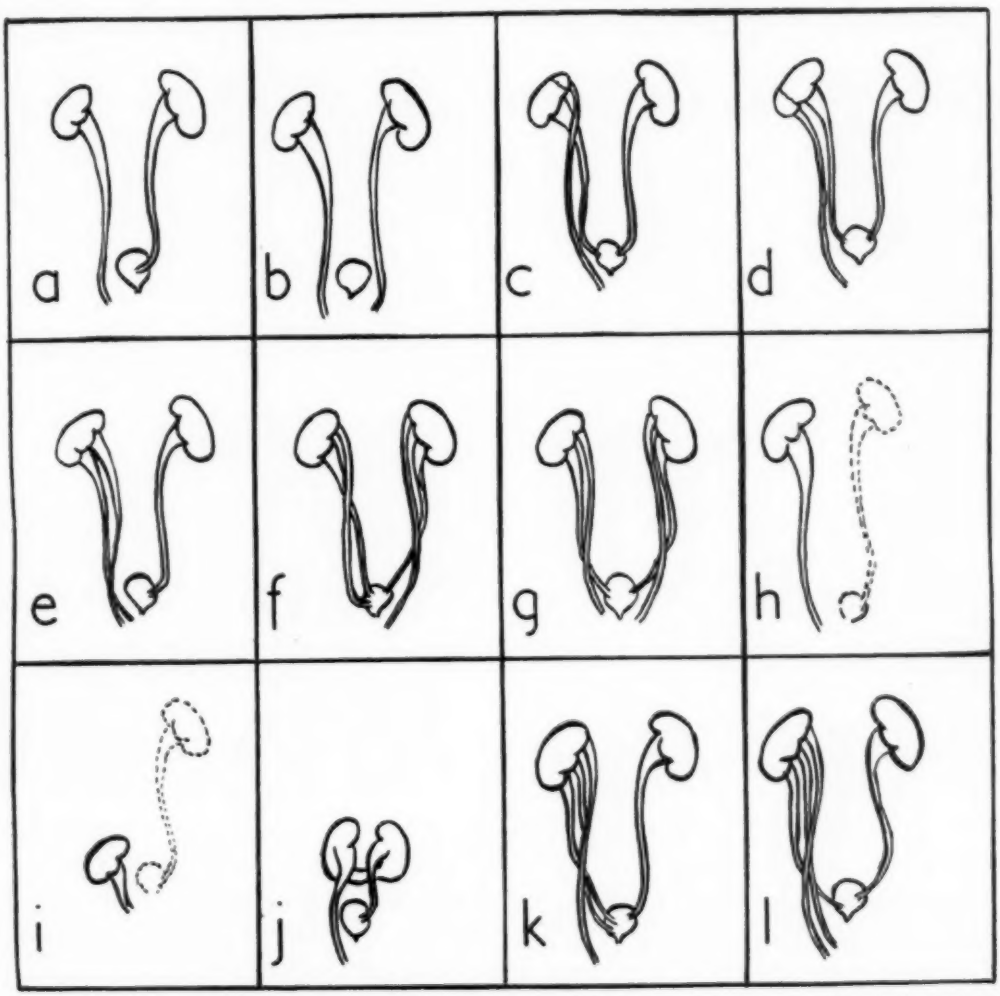

FIG. 393.—Associated upper urinary tract abnormalities. a, Unilateral single ectopic ureter; b, Bilateral single ectopic ureter; c, Unilateral double ureter with single ectopic orifice draining upper pole; d, Unilateral double ureter with single ectopic orifice draining lower pole; e, Unilateral double ureter with two ectopic orifices; f, Bilateral double ureter with one ectopic orifice; g, Bilateral double ureter with bilateral single ectopic orifices; h, Solitary kidney with single ectopic ureter; i, Solitary pelvic kidney with single ectopic ureter; j, Horseshoe kidney with single ectopic ureter; k, Unilateral triplicate ureter with homolateral single ectopic orifice; l, Unilateral triplicate ureter with homolateral double ectopic orifices.

In these cases it is probable that the vagina acts similarly, particularly if the orifice opens within or close to it.

Certain other anomalies of micturition may be present even in the absence of complicating pathology.

As a result of voluntary effort on the part of the patient, contractions of the pelvic diaphragm or urethral musculature may effectively control incontinence for a variable period of time. This limited period of incontinence may be very misleading. Innes Williams (1954) has also pointed out that urgency and frequency may be present in the absence

difference being more marked the greater the distance from the usual position. Dilatation therefore occurs commonly with subsequent infection. The latter is also favoured in females, since the orifice often opens on to surfaces which are readily contaminated.

DIFFICULTIES IN DIAGNOSIS

A full urological examination should be carried out whenever the condition is suspected. Except in those cases where the ureter drains into a seminal vesicle, rectal examination is inconclusive. Cystoscopy may show no abnormality or there may be

absence of a ureteric orifice and the associated hemi-trigone. Urethroscopy may reveal the orifice or some evidence of urethritis. Careful inspection in the female of the external urinary meatus, vestibule, vulva, vagina, and cervix is mandatory. However prolonged and careful this examination may be, it is often quite impossible to locate the minute orifice among the numerous crypts and folds. Every suspicious site should be probed and watch kept for the appearance of moisture in a previously dried area. Only visualization of the orifice coupled with catheterization and dye injection can prove conclusively the presence of the ectopic ureter and the associated upper urinary tract anatomy.

In some cases a ridge may be palpable in the vagina, thus affording some indication of the likely site of the opening. Vulval or vaginal excoriation may also be present owing to the urinary leak.

Intravenous Pyelography.—Reference to the various anomalies of the upper urinary tract known to be associated with ureteral ectopia reveals the necessity of a good pyelogram. Provided satisfactory function is present throughout the whole renal tissue, adequate contrast will be obtained.

The commonest renal anomaly, however, is the presence of a small element drained by the ectopic ureter and situated at the upper pole of the kidney. In many of these cases the concentrating power of this element is poor and little or no contrast is obtained. Very great care is therefore necessary in interpreting these films. The renal outline may be large in comparison with the pelvis and caliceal system or there may be a deficiency in the number of calices as compared with the opposite side. Minor points which may help in a doubtful case are slight lateral deviation of the upper pole of the kidney or lateral displacement of the uppermost visible calyx. Many cases, however, are indistinguishable from a normal pyelogram.

Complete absence of secretion on one side does not mean that there is no renal tissue present, as this may be hypoplastic, non-functioning, or in an abnormal situation.

FIG. 394.—*Case* 1. Intravenous pyelogram showing double pelvis on right.

FIG. 395.—*Case* 1. Retrograde pyelogram after catheterizing ectopic orifice.

Chromocystoscopy and Instillation of Dye into the Bladder.—When clinical suspicion of an ectopic ureter cannot be confirmed by the preceding investigations the following tests may be carried out :—

Instillation of methylene blue into the bladder followed by inspection of the vagina for staining, or delayed observation of a loose pack left in the vagina, will usually exclude the presence of a small vesico-vaginal fistula.

The second procedure, chromocystoscopy, is not often of great value since the renal element which is unable to concentrate radiopaque media also produces minimal concentration of dye. Persistent wetness from apparently clear fluid in the presence of deep blue bladder urine is, however, further evidence of an extravesical leak.

TREATMENT

Where full information regarding the upper urinary tract has been obtained, the method of treatment best suited to the case can be planned.

If, in spite of repeated investigations, neither the ectopic orifice nor the position of the associated renal element can be defined, a laparotomy should be advised.

It would seem wise in these circumstances to use a vertical subumbilical incision which can be extended to either flank if required. The bladder should be exposed extraperitoneally and both normal and ectopic ureters sought in the region of the bifurcation of the common iliac arteries. Subsequent dissection will then enable the anatomy to be determined and appropriate treatment carried out.

FIG. 396.—*Case* 2. Intravenous pyelogram showing double kidney on left.

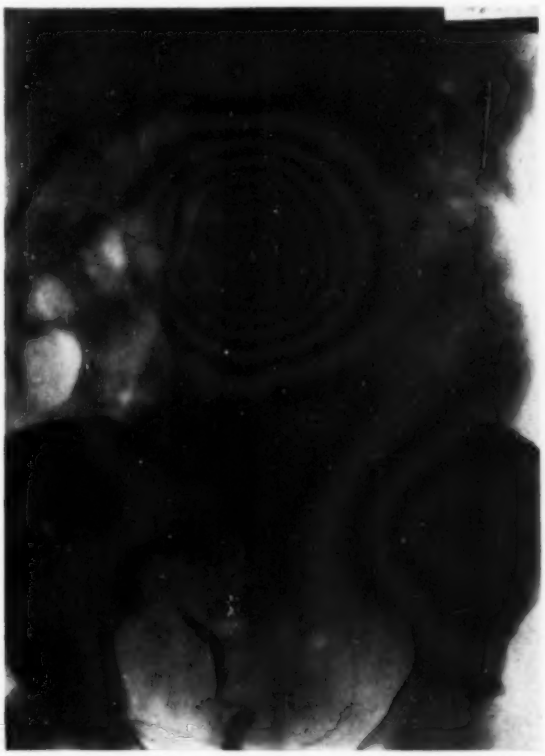

FIG. 397.—*Case* 2. Retrograde pyelogram after catheterizing ectopic orifice shows this to be associated with the right kidney.

The procedures which have been described are as follows :—

1. Ligation of ectopic ureter.
2. Transplantation of ureter into bladder or bowel.
3. Partial nephrectomy.
4. Partial nephro-ureterectomy.
5. Nephro-ureterectomy.
6. Uretero-ureterostomy.
7. Uretero-pyelostomy.

Each of these procedures may have its place in the individual case, although the advisability of the last two is very dubious.

There is as yet no general agreement on the correct method of treatment for the commonest abnormality, where the ectopic ureter drains the upper pole of an otherwise normally functioning kidney. In these cases choice lies between simple ligation of the ectopic ureter and excision of the upper pole of the kidney with or without the associated ureter.

In the absence of infection or dilatation, simple ligation is often satisfactory and a plea has been made for its use (Andrews and Vernon, 1943). The theoretical but as yet dubious relationship between hypertension and non-functioning renal tissue might be considered a contra-indication to this treatment.

The majority of cases, however, are best treated by resection of the small renal element together with the whole of the accessible ureter. Removal of the whole kidney is unnecessarily drastic since it is usually easy to define the segment of renal tissue to be removed either before or after ligation of its vascular pedicle.

It is my belief that the whole ureter should be removed when it is dilated or infected, since there are several cases on record where a difficult secondary ureterectomy has been required for persistent infection.

Although little trouble arises from the residual normal ureter following nephrectomy, the ectopic ureter cannot be placed in the same category, particularly when it opens onto the surface or into the female genital tract. The added risk of subsequent ascending infection must be borne in mind and this would seem to be an adequate reason for advocating total ureterectomy as a routine.

CASE REPORTS

Case 1.—Miss R. W., age 15 years, admitted to the Royal Free Hospital under the care of Mrs. D. M. S. Barclay. Adopted, aged 4½ years. At that time enuresis. Foreign body removed from vagina without any improvement. Never completely dry. Enuresis persisted, aggravated by coughing and laughing. Sleeps well. Frequency, D/N = 8/2. Psychiatric advice had been suggested. On

examination, nil abnormal except urinary leak from external meatus on straining. Vulva moist with some excoriation. Intravenous pyelogram, double kidney on right with ureter visible as far as iliac crest (*Fig.* 394).

May 29, 1951: Cystoscopy and retrograde pyelogram: ectopic orifice on posterior lip of urethral opening near midline. Clear efflux. Cystoscopy normal. C.S. bladder urine sterile. Specimen from ectopic orifice

FIG. 398.—*Case* 3. Intravenous pyelogram showing large left kidney. No secretion on right side.

FIG. 400.—*Case* 3. Operation specimen showing hypoplastic right kidney.

contained a few pus cells. Catheters passed 10 cm. up ectopic orifice and right ureteric orifice. Dye injection confirmed double kidney on right with upper pole draining through ectopic orifice (*Fig.* 395).

June 5: Hemi-nephro-ureterectomy. Separate artery to upper pole. Dry since operation.

Case 2.—Mrs. F. G., age 51 years, admitted to the Royal Free Hospital under the care of Mrs. D. M. S. Barclay. Complained of hernia for ten years, no longer controlled by truss. On examination large reducible L.I.I.H. Noticed to be wet. Volunteered information that she had been taken to another hospital as a child on account of this, but her parents had been told that nothing could be done about it. Ectopic orifice visible behind and to right of external meatus. Intravenous pyelogram, double kidney on left, apparently normal on right (*Fig.* 396). Dye injection via ectopic orifice showed this to drain upper pole of right kidney (*Fig.* 397). Cystoscopy revealed only two ureteric orifices. All urine specimens sterile.

March 31, 1953: Hemi-nephro-ureterectomy. Upper pole cystic with separate vessels. Dry since operation.

Case 3.—Miss S. R., age 22 years, admitted to King's College Hospital under the care of Mr. J. G. Yates-Bell. Complained of urinary leak since birth, plus normal micturition. Conscious of normal bladder distension. Occasionally dry through the night. Normal twin sister.

On examination, N.A.D. apart from labial excoriation, a small quantity of watery fluid in the vagina and a palpable ridge running down the right vaginal wall.

FIG. 399.—*Case* 3. Cystogram showing bladder disposed mainly to the left side.

Intravenous pyelogram showed a large left kidney secreting normally, a small bladder disposed mainly to the left of the pelvis, and no evidence of secretion from a right kidney (*Figs.* 398, 399). Cystoscopy revealed absence of the right ureteric orifice and associated hemi-trigone. Urethroscopy normal. No ectopic orifice could be detected. Intravenous indigo carmine produced no extravesical staining. Uriodone injected into the cervical canal gave a normal hysterogram. C.S. urine and blood-urea normal.

Discharged home to await repeat investigations. Developed pyelitis. Re-admitted two months later. Repeat investigations confirmed previous findings. Methylene blue instilled into bladder—no vesicovaginal fistula.

AT OPERATION—Oct. 25, 1956: Midline subumbilical incision. Right ureter found crossing a small right internal iliac artery. Traced down to below bladder, and upwards to a small piece of renal tissue situated just above level of umbilicus. Methylene blue injected into ureter appeared in lower part of vagina. Excision of hypoplastic right kidney and ureter (*Fig.* 400). Dry since operation.

Case 4.—Miss H. H., aged 7½ years, admitted to Bromley Hospital under the care of Mr. W. P. Greening. Complained of incontinence since birth. Not affected by posture. Normal twin brother. Vaginal calculus removed November, 1949. Ectopic ureter suspected but not proven (Eton, 1954). Treated as functional. Developed pyelitis and was then referred to Mr. Greening.

Intravenous pyelogram, Feb. 2, 1950: Lumbarization of S.1 and spina bifida of S.1, 2, 3. Additional shadow above upper pole of left kidney (*Fig.* 401). Left hemi-nephrectomy with excision of upper ureter. Separate vessels to firmly adherent upper pole. Dry since operation. Subsequently ectopic orifice in left vaginal fornix catheterized (*Fig.* 402) and residual ureter removed.

Case 5.—Miss R. T., age 19 years, admitted to the Royal Marsden Hospital under the care of Mr. D. H. MacLeod. Complained of incontinence of urine since birth. Occurred day and night; aggravated by exertion. Considered to be psychological. Anterior colporrhaphy performed elsewhere without improvement.

On examination (Nov. 22, 1954): No apparent abnormality. No vesico-vaginal fistula. Cystoscopy normal. Intravenous pyelogram and bilateral retrograde pyelogram reported normal (*Figs.* 403, 404). Nov. 29: Small opening, from which a gush of urine appeared, seen on anterior vaginal wall 1 cm. from introitus.

FIG. 401.—*Case* 4. Intravenous pyelogram showing additional shadow above left renal pelvis.

FIG. 402.—*Case* 4. Showing residual ureter after nephrectomy. Subsequently excised.

FIG. 403.—*Case* 5. Intravenous pyelogram showing blunting and slight lateral deviation of right upper calyx.

FIG. 404.—*Case* 5. Retrograde pyelogram confirms abnormal right upper calyx.

Opening catheterized and retrograde injection showed track passing up right side of abdomen (*Fig.* 405).

At operation a dilated ectopic ureter was traced towards the posterior fornix. No renal tissue palpable at upper end. Ureter excised as high as possible to point

FIG. 405.—*Case* 5. Retrograde pyelogram after catheterizing ectopic orifice.

where it crossed the normal right ureter below. Dry since day of operation.

Case 6.—Mrs. N. D., age 40 years, admitted under the care of Mr. C. K. Vartan. Complained of stress incontinence. A poor witness who gave a history of some incontinence as a child.

Colporrhaphy had been performed to relieve the stress incontinence. A year later this was repeated as the first operation had not proved satisfactory.

During the second operation a small tunnel was opened just to the right of the urethra. A probe was passed along this channel and contrast medium injected. This revealed a tubular structure running up to the right loin. Immediate laparotomy was performed and this was found to be an accessory ureter which terminated in a knob of renal tissue just lateral to the right kidney. The middle six inches of this ureter were excised. Subsequently an intravenous pyelogram showed normal functioning kidneys on both sides. Complete cure of the incontinence was achieved after performing a sling operation at a later date.

SUMMARY

1. From the world literature it has been possible to collect 459 cases of extravesical ectopic ureter, having a total of 494 ectopic orifices.

2. Six additional cases are reported.

3. The embryology is discussed and a possible explanation for some of the anomalies suggested.

4. The sites of the ectopic orifices and the associated upper urinary tract anomalies are described.

5. The classical symptomatology and its variations are discussed.

6. The necessity for full investigation of the urinary tract is stressed, and the treatment briefly considered.

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TUBERCULOUS ULCERATION AS A CAUSE OF PERFORATION OF THE DISTAL DUODENUM

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WHILST perforation commonly occurs in the first part of the duodenum it is rare in the more distal parts. In such cases it usually complicates an ulcer which is tuberculous in nature. The following case is presented as an example of this unusual condition.

CASE REPORT

A 53-year-old spinster was referred to the Out-patients Department on Nov. 15, 1954. She gave a 3-months' history of loss of appetite, abdominal pain, and intermittent diarrhœa. The anorexia had become progressively worse with consequent loss of weight and increasing weakness. The pain, which was unrelated to meals, was situated in the right iliac fossa and the hypogastrium. During the attacks of diarrhœa the stools were yellowish in colour, but never contained mucus or blood. For many years she had vomited small amounts of frothy fluid after meals, but this had not been worse since the onset of the other symptoms. She had never felt really well since having had rheumatic fever in 1935. There was no history of any previous illness apart from this.

Examination revealed a thin, undernourished woman. A small, slightly tender mass was felt in the right iliac fossa. It was difficult to define, but gave the impression that it was fixed in position and was not fluctuant. No other abnormality was found in the abdomen. Rectal examination was negative. The heart was slightly enlarged and the blood-pressure was 165/90. The cardio-vascular system was otherwise normal. No abnormalities were found in the other systems. Examination of the blood revealed a hæmoglobin concentration of 70 per cent and an erythrocyte sedimentation rate of 76 mm. in the first hour.

The case was provisionally diagnosed as ' ? Carcinoma of the cæcum ' and arrangements were made to admit her for further investigation in a few days' time. That night, however, she developed severe abdominal pain associated with a rigor. Six hours after the onset of the pain she passed a large quantity of dark blood per rectum. She was admitted as an emergency. By then the severe pain had been replaced by a steady ache in the lower abdomen. On examination she was found to be mildly shocked. The pulse-rate was 115 per minute and the blood-pressure had fallen to 115/65. The mass in the right iliac fossa had enlarged to the size of an orange and was very tender. It could be moved slightly in a plane at right angles to the line of attachment of the small bowel mesentery. There was no rebound tenderness or hyperæsthesia. The bowel sounds were present and normal in character. The hæmoglobin concentration was now found to be 52 per cent and the white-cell count was 28,500 per c.mm. The temperature was normal on admission but rose to 99·6° F. in a short time. A transfusion of whole blood was started, but despite this the pulse-rate rose steadily. In view of these findings it was decided to perform a laparotomy.

FIRST OPERATION.—A right para-umbilical incision was made. No free fluid was present in the peritoneal cavity. A fluctuant swelling was found in the root of the mesentery of the small intestine. The whole of the small and large bowel contained dark blood. The stomach, first and second parts of the duodenum, and the pancreas appeared to be normal. The lesser sac was opened but nothing abnormal was found. A wide-bore needle was inserted into the swelling but aspiration failed to withdraw anything. The swelling was then opened by blunt dissection and a large necrotic cavity entered. A finger passed into this was directed upwards and entered the third part of the duodenum at its junction with the fourth part. After a specimen of the necrotic tissue had been taken for pathological examination the opening in the peritoneum was closed and covered with a patch of omentum. The second and third parts of the duodenum were mobilized medially and the third separated from the superior mesenteric vessels. This revealed an opening on the inferior border of the third part of the duodenum. This opening communicated with the cavity in the root of the mesentery and its margins were bleeding quite freely. The ragged edge was excised and a transverse closure obtained. A drain was left down to the necrotic area and the abdomen was closed.

POST-OPERATIVE PROGRESS.—Penicillin and streptomycin were given during the first week after operation. The initial progress was satisfactory and it was possible to discontinue the intravenous drip and gastric suction on the fourth day. Three days later, however, large quantities of bile and pancreatic secretions began to issue from the drain. This continued for two weeks, after which it ceased abruptly. During this period fluids and electrolytes were given intravenously. An indurated area appeared in the right loin during the third week and was accompanied by a rise in temperature. This rapidly responded to a second course of streptomycin. Ten days later the inflammation in the loin flared up again and on this occasion was associated with mental confusion and acute cardiac failure. Digoxin and aureomycin controlled these complications. Thereafter the patient made uneventful progress and was discharged nine weeks after the operation.

During the whole of the convalescent period she continued to vomit frothy fluid after meals. Radiological examination before her discharge from hospital showed that this was due to cardiospasm. Some delay in gastric emptying was noted, but no abnormality was seen in the duodenum. Dilatation of the cardia was begun two months later, but as the results were not satisfactory a Heller's operation was performed in June, 1955. Since then she has had no further vomiting.

PATHOLOGY.—Histological examination of the specimen of necrotic tissue showed that it was composed of organized granulation tissue.

Examination of the tissue excised from the margins of the perforation showed that it was duodenal wall with some fat necrosis and a non-specific inflammatory reaction in the deeper layers. The mucosa was heavily infiltrated with lymphocytes and some follicular hyperplasia of lymphoid tissue was seen. The appearances were those of a non-specific chronic duodenitis.

In view of these histological findings the cause of the perforation was thought to be either peptic ulceration or inflammation of a diverticulum. Both conditions are extremely uncommon in the third part of the duodenum and no account of perforation complicating either of them in this site could be found in the literature.

FURTHER HISTORY.—Three months after the Heller's operation the patient was again referred to the hospital complaining of malaise and pain over the sacrum which had been present for five weeks. One week after the onset of these symptoms she had developed diarrhœa which had persisted thereafter. There had been no melæna. Apart from occasional cramp-like pains in the lower abdomen she had no other symptoms.

Examination revealed the presence of a small, tender mass in the right iliac fossa and some tenderness in the

ght loin. On rectal examination a tender mass was palpated high up on the right side. No other abnormalities were found.

The patient was re-admitted to the medical unit where blood- and stool-cultures, Paul Bunnell, and Widal reactions were found to be negative. The hæmoglobin concentration was 59 per cent and the white-cell count 7,000 per c.mm. The temperature was variable over the range 98° F.–101° F. The hæmoglobin concentration was raised to 90 per cent by blood transfusion and a course of terramycin was started. There was some improvement in the general condition, but as the abdominal signs remained unchanged, a further laparotomy was performed on Oct. 28, 1955.

SECOND OPERATION.—The abdomen was re-opened through the old paramedian wound. A granulomatous lesion was found between the abdominal wall and the second part of the duodenum and was excised. It appeared to be the site of the drain used in the first operation. A small white mass was found lying anterior to the lower pole of the right kidney and this was also removed for examination. In the ileo-cæcal region was a mass of caseating glands which had progressed to abscess formation. As it was not possible to free the mass from the cæcum and ascending colon, a hemicolectomy with ileo-transverse anastomosis was carried out.

The patient made an uneventful recovery from this operation and was then transferred to a tuberculosis unit.

PATHOLOGY.—The first two specimens consisted of fibro-fatty tissue containing areas of fibroblasts amongst which were lymphocytes, endothelial cells, and a few giant cells with small areas of caseous material. The appearances were those of tuberculous granulation tissue.

The hemicolectomy specimen consisted of 20 cm. of colon including the cæcum to which was attached 4 cm. of ileum. The surrounding tissues were matted together and fibrotic. At a point 5 cm. from the ileo-cæcal valve there was a discrete perforation of the cæcum. The appendix showed no thickening of its wall. Sections taken from the mesentery consisted of tuberculous granulation tissue in which there was some caseation. Sections through the appendix showed no histological abnormality. A section through the site of perforation of the cæcum showed an area of ulceration in the mucosa. In the wall of this and in the serosa were numerous capillary channels lying amongst delicate fibro-fatty tissue which was heavily infiltrated with lymphocytes. A small sinus tract was seen surrounded by necrotic material and histiocytes. There was no specific evidence of tuberculosis in this section through the ulcer of the cæcum.

In view of these findings the biopsy specimens obtained at the first operation were re-examined. After a careful search a few poorly formed giant cells were made out in the necrotic areas. No evidence of true caseation was discovered. It appeared that what had been taken to be inflammatory debris at the original examination was, in fact, tuberculous granulation tissue.

DISCUSSION

Aetiology.—Tuberculosis of the duodenum may occur either as a primary lesion or secondary to a focus of infection elsewhere in the body. Primary lesions are uncommon and usually form part of an infection which affects the stomach and duodenum simultaneously. They are probably due to infection by ingested bacilli. Matthews, Delaney, and Dragstedt (1932) noted that duodenal lesions unassociated with generalized tuberculous infections often have symptoms extending over a long period whilst the lesion itself shows histological evidence of recent acute inflammation. This led them to suggest that such primary lesions are due to tuberculous infection superimposed on a chronic peptic ulcer. In these cases the tuberculous nature of the lesion is seldom suspected before operation or autopsy. Similar views had been expressed in an earlier paper by Schwatt and Steinbach (1923).

It is also possible that duodenal tuberculosis may result from the direct spread of infection from nearby lymphatic glands. Ostrum and Sorber (1948) favour this manner of infection in view of their findings that in nearly all the recorded cases of gastroduodenal tuberculosis enlarged mesenteric and retroperitoneal glands were found. Previously all such infected nodes had been attributed to spread from a primary lesion in the duodenum, but it seems likely that, in some cases at least, the glands were infected from the small intestine and those lying in proximity to the duodenum became adherent to it and thus allowed the infection to enter its wall.

Duodenal tuberculosis occurs much more frequently as part of a generalized infection. Most reported tuberculous duodenal ulcers were incidental findings at autopsy in patients who had died of the advanced disease. Few cases had any symptoms referable to the duodenal ulceration during life although, in some cases, such symptoms may have been masked by those due to coexistent ulceration of other parts of the alimentary tract. Migliaccio (1946) reported that up to then less than a dozen cases of duodenal tuberculosis occurring as solitary lesions and giving rise to symptoms had been reported.

Incidence.—Fenwick and Dodwell (1892) reviewing 500 cases in which tuberculosis of the alimentary tract with ulceration was present at autopsy noted that the duodenum was affected in 3·4 per cent of them. None had tuberculosis of the duodenum as a primary lesion. Perforation had occurred in 10 per cent of the duodenal ulcers, an incidence at least ten times greater than that found in ulcers of other parts of the alimentary tract. The ulcers were most common in the distal parts of the duodenum, but in the 4 examples of perforated duodenal ulcer in this series and in a series they collected from the literature, 3 were in the first part. This suggests that the gastric secretions may be an important factor in causing a tuberculous ulcer to perforate.

The incidence given by other authors differs from that given above. Perry and Shaw (1894) found 11 examples of tuberculous duodenal ulceration in 17,652 autopsies performed over a period of sixty-six years. Gossman (1913) found 5 such cases in 2360 autopsies on patients who had died of tuberculosis, whilst Schwatt and Steinbach found 3 examples amongst 199 similar patients.

Perforation of such an ulcer is rare and, including those mentioned above, only 15 cases appear to have been recorded in the literature.

Pathology.—The duodenal lesions take the form of typical tuberculous ulcers which may be single or multiple. In those situated in the first part of the duodenum there may be evidence that the tuberculous infection is superimposed on a chronic peptic ulcer. In rare cases the infection may be of the hyperplastic type not uncommonly seen in the ileo-cæcal region (Matthews and others, 1932; Garvin, 1930). In most cases tuberculous ulceration is found lower down the alimentary tract. Enlargement of the

mesenteric and retroperitoneal lymph-nodes is an almost constant feature. When perforation into the peritoneal cavity occurs it is usually walled off by previously formed adhesions with the formation of an abscess in the sub-hepatic region. Perforation may occur into the retroperitoneal tissues and the resulting abscess then tends to track towards the cæcum. One case is recorded in which such an abscess ruptured into the cæcum (Rintel and Schultzen, 1869). Usually these abscesses take a long time to form. Perforation may occur into an adherent viscus or into a vessel such as the aorta with rapidly fatal results (Beresford-Jones, 1941).

Symptoms.—The great majority of reported tuberculous duodenal ulcers occurred in the terminal phases of advanced tuberculosis. Matthews and others collected 123 cases from the literature and found that only 18 of these had produced symptoms referable to the condition.

Melchior (1917) found that the majority of tuberculous ulcers which cause symptoms are situated in the first part of the duodenum, whereas it had been noticed by other authors (Ostrum and Sorber; Fenwick and Dodwell) that tuberculous ulcers in general are most common in the distal parts. These findings suggest that the symptoms are due to the effects of the gastric secretions on the ulcer.

The main symptom is upper abdominal pain. This is of typical duodenal ulcer type in the majority of cases, but in some may be felt after meals or be quite unrelated to them.

Vomiting often accompanies the pain and usually relieves it.

Diarrhœa is found in cases with coexistent ulceration of the intestines.

Perforation is accompanied by an increase in the pain. Nausea and vomiting follow, together with hæmatemesis or melæna in most cases. The pain tends to remain localized owing to the perforation being walled off by adhesions.

Symptoms of generalized tuberculosis may be found in those patients in whom the condition has not been already diagnosed.

Signs.—Tuberculous lesions of the duodenum rarely give rise to any local physical signs. In a few cases there is vague tenderness over the upper abdomen. There are usually signs of chronic ill-health and, possibly, tuberculosis elsewhere in the body. In some cases this has taken the form of a peripheral lymphadenopathy.

Barium meal examination may demonstrate a lesion in the duodenum, but this is usually mistaken for peptic ulceration when it occurs in the first part and for a neoplasm when in the more distal parts. The diagnosis in these cases is made on pathological examination of the specimen removed at the subsequent laparotomy.

When perforation occurs the resulting abscess can be palpated as a lump in the abdomen. It may be in the sub-hepatic position or along the line of attachment of the small bowel mesentery. It is usually fixed and is elastic in consistency. It may or may not be tender, depending on the degree of inflammation set up in the surrounding tissues.

Treatment.—Correct treatment depends on correct diagnosis, and the absence of the latter has been a feature of the cases reported up to now. In the majority no treatment had been given for the duodenal condition as it had not been recognized until autopsy. In the others the lesions were regarded as peptic ulcers or neoplasms and were treated accordingly. In the first group the finding at operation of a hard mass related to the duodenum led to a revision of the diagnosis and, under the impression that a tumour was present, a partial gastroduodenectomy was performed. In the second group the usual treatment of the lesion was a resection of the lesion followed by gastrojejunostomy. In one case treated by partial gastrectomy a tuberculous ulcer in the duodenal stump perforated and caused the death of the patient five weeks after the operation (Ostrum and Sorber).

Matthews and others suggested that the choice of treatment in those cases in which tuberculosis was confined to the duodenum lay between radical resection and gastrojejunostomy. When tuberculosis is present elsewhere in the body it would seem inadvisable to attempt to treat the duodenal lesions surgically, unless they were complicated by perforation, giving rise to severe symptoms, or could not be differentiated from non-tuberculous conditions requiring surgical treatment. In all other cases treatment should be along established lines by sanatorium régime and the administration of anti-tuberculosis drugs.

Perforation should be treated by evacuation of the resulting abscess and closure of the perforation after excision of its diseased margins. Drainage is advisable in view of the potentially diseased nature of the duodenal wall and the consequent danger of poor healing and leakage. The appropriate anti-tuberculosis drugs should be given during the post-operative period, preferably after consultation with the tuberculosis physican who will be responsible for the subsequent treatment.

SUMMARY

1. A case of perforation of a tuberculous ulcer of the third part of the duodenum which presented with abdominal pain and massive intestinal hæmorrhage is described.

2. The aetiology, pathology, clinical features, and methods of treatment are discussed.

3. The difficulties of diagnosis are stressed.

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PORTAL PYÆMIA

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PORTAL pyæmia or pyogenic liver abscess consequent upon a primary focus of inflammation within the territory drained by the portal venous system is a condition which does not lack description in the medical literature of the last hundred years. It was and still is an uncommon condition, but one which has been reported sufficiently often for its features to become well recognized. The problems that have attended the condition have been those of early diagnosis, of treatment, and of high mortality.

During the 5-year period 1951–6, 10 verified cases of multiple pyogenic liver abscess have been seen in this hospital. In only 4 cases was a correct diagnosis made during life, and in 3 patients the abscesses were drained; of the 10 patients, one alone survived.

It is the purpose of this paper to present briefly the case-histories of 2 patients in whom the abscesses were drained, to tabulate the principal features of the remaining 8, and to emphasize that even in this era of antibiotics and enhanced aids to surgical diagnosis, the problem of portal pyæmia remains much as it was twenty or more years ago.

CASE REPORTS

Case 1.—Mr. W. S., a gardener, aged 46, was admitted to hospital on Jan. 14, 1954, complaining of shivering attacks and pain in the right hypochondrium for seven days. There had been no vomiting and no alteration of his normal bowel habit and he would not admit to either a cough or any urinary symptoms. He had never been abroad nor had any previous illnesses.

On admission the patient looked ill, with a temperature of 102° F. A full examination revealed tenderness and rigidity in the right hypochondrium. The liver was not palpable and there was no evidence of jaundice. A preliminary diagnosis of acute cholecystitis was made. Two days after admission a faint tinge of jaundice became apparent and remained so for one week before disappearing. Bile was present in the urine. Repeated daily chills and rigors began to occur and the temperature chart showed an intermittent fever.

A white blood-count, on admission, was 23,000 per c.mm. with 83 per cent neutrophil polymorphs. Radiographs of the chest and abdomen were normal. Repeated blood-cultures were negative, as were agglutination tests for enteric fever, brucellosis, and Weil's disease. Albumen was present in the urine.

On admission, penicillin treatment 500,000 units b.d. was begun. After one week, streptomycin 0·5 g. b.d. was also given. No clinical improvement occurred and the pyrexia continued. Four weeks after admission, œdema of the skin over the lower right ribs was noted, and laparotomy performed with a pre-operative diagnosis of empyema of the gall-bladder.

AT OPERATION (Mr. N. C. Tanner).—Through a right paramedian incision, the liver was found enlarged and needling it revealed multiple abscesses in the left lobe, but none in the right. The sigmoid colon was the site of acute diverticulitis; the rest of the viscera were normal.

After packing the liver off from the general peritoneal cavity with a gauze roll, the abscesses were opened, and rubber drains inserted down to them were brought out through the abdominal wound. A diagnosis of portal pyæmia from diverticulitis coli was made.

Culture of the pus obtained from the liver abscesses grew *B. coli* sensitive to streptomycin, chloramphenicol, and terramycin.

Post-operatively the pyrexia dropped for a week as pus drained, but thereafter rigors and a continuous fever recurred despite the administration of antibiotics. *Proteus vulgaris* was now cultured from the wound. Six weeks after operation, the patient developed signs of a cerebral abscess and after a gradual decline in his condition, died on April 24, 1954, before neurosurgical aid could be summoned.

AT AUTOPSY.—The relevant findings (confirmed histologically) were diverticulitis of the sigmoid colon together with multiple abscesses, some old and some recent, in both right and left lobes of the liver. These abscesses were in relation to the branches of the portal vein. A left cerebral abscess was present but no pyæmic abscesses were found in any other organs.

Comment.—In this patient, despite drainage and the giving of antibiotics, death occurred. The delay in diagnosis was undoubtedly a factor contributing to death. The fact that at autopsy more abscesses were discovered in the liver means either that they were present but undiscovered at time of operation, or that they developed later. In view of the recurrence of rigors and fever after operation, it is likely that more abscesses developed during the post-operative period either by embolic spread from the primary focus in the colon or by spread within the liver from the abscesses already present at time of operation.

Case 2.—Mr. C. F., a secretary, aged 54, was admitted to hospital on April 16, 1956, for investigation of a pyrexia of unknown origin of 10-days' duration, together with rigors. He had no other complaints and would not admit to any bowel symptoms. He had previously been in good health, and had never been abroad.

On admission he looked well and, apart from a fever, no abnormality could be found on clinical examination.

W.B.C. were 28,000 with 80 per cent polymorphs. The urine contained albumin; agglutination tests for enteric fever and brucellosis were negative, as were repeated blood-cultures. Liver function tests showed no evidence of jaundice and were normal. Radiographs of chest and cholecystogram were normal.

Following admission an intermittent fever developed and rigors and chills were repeated often, despite treatment with penicillin 500,000 units b.d. and streptomycin 0·5 g. b.d. Seventeen days after admission, the liver became palpable and tender and a faint jaundice appeared. (Urine contained bile; serum bilirubin 3·4 mg. per cent.) A diagnosis of pylephlebitis was made and on May 3, 1956, a laparotomy was performed.

AT OPERATION (Mr. N. C. Tanner).—As before, a right paramedian incision was made and the liver was found enlarged. The sigmoid colon was once again the site of acute diverticulitis. After packing the liver off with a gauze roll, the liver was needled and two abscesses were discovered in the right lobe, but none in the left. These were opened and rubber drains placed down to them.

A separate incision was made in the left iliac fossa and the sigmoid colon was exteriorized over a glass rod and the inferior mesenteric vein ligatured in continuity.

Three days later, the exteriorized colon was excised and the ends left as a colostomy.

Post-operatively the patient's temperature rapidly returned to normal and an uneventful recovery was made. The gauze roll was removed on the tenth day, and after three weeks pus ceased to discharge from the liver. An extraperitoneal closure of the colostomy was performed and on July 12, 1956, the patient left hospital and has so far remained well.

chills and rigors are few and far between (Rothenberg and Linder, 1934; MacFadzean, Chang, and Wong, 1953). However, although chills and rigors were repeated daily, they tended to fade from the illness after a week or ten days; this occurred most noticeably in *Cases* 1, 2, and 7.

Of the 10 patients, 6 had a tender and palpable liver at some stage of their illness, together with dull

Table I.—SOME OF THE FEATURES OF

Case No.	Sex	Age	Primary Focus		Diagnosis of Portal Pyæmia made	Liver Lobes involved	Pus Elsewhere
			Site and Nature	Silent or Clinically Obvious			
1	M.	46	Diverticulitis of sigmoid	Silent	Operation	Right and left	None
2	M.	54	Diverticulitis of sigmoid	Silent	Clinically and operation	Right	None
3	M.	29	Unknown	—	Operation and autopsy	Right and left	Pancreatic abscess
4	M.	17 mth.	Appendicitis	Obvious	Autopsy	Right and left	General peritonitis
5	M.	64	Unknown	—	Autopsy	Right	Subphrenic abscess
6	M.	4 wk.	Umbilical cord	Obvious	Clinically and autopsy	Right and left	None
7	F.	29	Salpingitis and pelvic abscess	Obvious	Autopsy	Right and left	Subphrenic abscess
8	F.	70	Diverticulitis of sigmoid with peritonitis	Obvious	Autopsy	Left	Subphrenic abscess
9	M.	54	Diverticulitis of sigmoid	Silent	Autopsy	Right and left	Subphrenic abscess
10	M.	62	Diverticulitis of sigmoid	Silent	Autopsy	Right and left	None

Culture of the pus from the liver was sterile.

Histology of the colon resected showed the changes of acute and chronic diverticulitis with intramural abscesses.

Comment.—The diagnosis of liver abscess was correctly made without any clinical evidence of a primary focus of inflammation. In order to prevent embolic spread after operation, as had probably occurred in *Case* 1, the sigmoid colon was exteriorized and the inferior mesenteric vein ligated in continuity. This ligation prevented spread by emboli until the exteriorized bowel could be resected, as it was three days after operation.

DISCUSSION

Symptoms and Signs of Portal Pyæmia.—On considering the 10 cases shown in *Table I* no new symptom or sign can be found to add to what is already in the literature on portal pyæmia. Rather, one has to emphasize that the clinical picture is the same to-day as it was in the early years of the century.

The mode of presentation of patients with portal pyæmia is discussed later under diagnosis. All cases (except *Case* 4, who died within twenty-four hours of admission to hospital) ran an intermittent or a continuous fever with a daily variation of at least 2° F. If chills and rigors occurred, they did so repeatedly daily, thus indicating that the abscesses were multiple in the liver. This is in contrast to the patient with a solitary pyogenic liver abscess in whom pain in the right hypochondrium, in one case radiating to the right shoulder and, in two others, aggravated by breathing. Opinion on the frequency of occurrence of pain and a palpable liver has varied, but Ochsner, deBakey, and Murray (1938) in reviewing 830 cases of portal pyæmia collected from the literature, together with 47 of their own personal cases, found pain to be present in 90 per cent of the collected cases and in 91 per cent of their personal cases. A palpable liver was present in 89 per cent of the collected, and in 61 per cent of the personal, cases.

Jaundice occurred in less than half of the cases presented here. In 2 patients it was slight and transient; in 2 others it was deep and terminal. The frequency with which jaundice occurs appears to be related to the number of abscesses present, for in solitary pyogenic abscess it is very rare (MacFadzean and others). It can be expected in less than 50 per cent of cases with multiple abscess (Ochsner and others) and, when it persists and deepens, jaundice can then be taken to indicate a poor prognosis.

Malaise, anorexia, and loss of weight also occurred here in the same frequency as previously reported; ascites was present in only one case, as was œdema of the parietes overlying the liver.

In those cases where a W.B.C. count was performed, a leucocytosis of the order of 20,000 or more was found with a preponderance of polymorphs.

To this account must of course be added the symptoms and signs of the primary focus of inflammation, if it be present or manifest clinically.

The Primary Focus of Inflammation.—In portal pyæmia this may lie within any of the abdominal viscera. An interesting feature of the cases seen at this hospital is that appendicitis constituted this primary focus in only one case. Appendicitis has long been stressed as the commonest single cause of portal pyæmia—hence the coining of the term *le foie appendiculaire* by Dieulafoy (1898). The early reports of portal pyæmia, particularly by the French physicians of the late nineteenth century, were all concerned with it as a complication of appendicitis, and it was perhaps in this way that there arose an exaggerated view of the frequency with which portal pyæmia was due to appendicitis. Thus, of the personal cases of Ochsner and others in only 10·6 per cent was appendicitis the primary lesion.

What then is the position to-day? At the time of writing it is impossible to answer this question exactly, for I have been unable to find any reported series of portal pyæmia of sufficient size from which to draw a conclusion about the exact incidence of appendicitis causing portal pyæmia since the introduction of antibiotics.

Diverticulitis coli was the source of the liver abscesses in 5 of the 10 cases here; in 4 of these it was 'silent' clinically. Similar cases of 'silent' diverticulitis, with portal pyæmia, have been reported by Cooke (1937) and Dearlove (1954). This 'silence' of the initial focus of inflammation has not been sufficiently stressed in the past. Ochsner and others pointed out that in a large proportion of cases of portal pyæmia the primary focus is not found. To this group, the term cryptogenic liver abscess has been applied. Beaver (1931) considered that in these cases the primary lesion was within the territory of the portal system, but was of such a minor character as to pass unrecognized clinically. It may be that as seen here diverticulitis contributes a large proportion of these cryptogenic liver abscesses.

It is interesting that the development of portal pyæmia, and whether or not the abscesses be solitary or multiple, would seem to bear no constant relationship to the severity or the duration of the primary focus of inflammation. Thus, a patient with a very bad appendix phlegmon may not develop liver abscesses, whilst another, symptom-free with diverticulitis, will develop multiple liver abscesses and die. The conclusion to be drawn is that it is not necessary to have overwhelming clinical evidence of a primary focus of inflammation in order to make a diagnosis of portal pyæmia.

TEN CASES OF PORTAL PYÆMIA

Swinging Fever	Rigors and Chills	Pain over Liver	Palpable Tender Liver	Jaundice	W.B.C. Polymorphs	Antibiotics	Effect on Illness without Drainage	Drainage performed	Result
+	Repeated	+	+	Transient	30,000 (83 per cent)	Penicillin, streptomycin, aureomycin	None	Yes	Death
+	Repeated	+	+	Transient	28,000 (80 per cent)	Penicillin, streptomycin	None	Yes	Lived
+	Repeated	+	+	Absent	21,700 (86 per cent)	Penicillin, streptomycin, terramycin	None	Yes	Death
—	—	—	—	Absent	—	—	—	No	Death
+	Repeated	+	+	Absent	60,000 (93 per cent)	Penicillin, streptomycin, aureomycin	None	No	Death
+	?	—	+	Terminal	46,000 (55 per cent)	Aureomycin	Control—relapse later	No	Death
+	Repeated	+	+	Absent	18,600 (91 per cent)	Penicillin, streptomycin, aureomycin	None	No	Death
+	?	+	+	Absent	18,800 (83 per cent)	Penicillin, streptomycin	None	No	Death
+	Repeated	+	+	Terminal	26,000 (95 per cent)	Penicillin, streptomycin	None	No	Death
+	?	+	+	Absent	—	Penicillin	None	No	Death

Association of Subphrenic with Liver Abscess.—Four of the 10 cases in *Table I* had associated subphrenic abscesses. However, in none of these 4 was a direct communication demonstrable at autopsy between the subphrenic and the liver abscesses. It is therefore probable that in each case they arose separately. Harley (1955) found that of 188 patients with subphrenic abscess, 11·2 per cent had also an abscess within the liver. He concluded that when such abscesses occurred in the same patient, they most often arose secondary to a primary focus within the abdomen also. Whereas a liver abscess may, by direct spread, give rise to a subphrenic abscess, the converse is very rare.

This association has a practical significance in that the finding of a subphrenic abscess may distract the surgeon's attention, and liver abscesses may thus not be found as in *Case* 7. If, therefore, the clinical picture suggests hepatic abscess, the presence of subphrenic pus should not prevent needling of the liver from being performed.

Diagnosis.—The facility with which this will be made correctly will depend to a large extent on the mode of presentation of the patient, and whether

or not the surgeon has seen a case of portal pyæmia before, or is sufficiently aware of the condition. To some extent geography plays a part here, as undoubtedly in an area where liver abscess is not uncommon—e.g. Hong Kong (MacFadzean and others)—the diagnosis will be made more readily than in London.

The mode of presentation can be in one of three ways :—

1. Where the primary lesion is clinically obvious, and where signs of a pyæmic liver abscess supervene.
2. Where the primary lesion has been dealt with at operation and where, in the post-operative period, signs of liver abscess occur.
3. Where the primary lesion is ' silent ' clinically.

The principal features are those of chills and rigors, with a swinging pyrexia, a high W.B.C. count, and a tender, palpable liver, with or without slight jaundice. Radiology may be of value in diagnosis, if an elevated and immobile diaphragm is found, but was of no help in any of the 10 cases discussed here. Subphrenic abscess, lobar pneumonia, empyema of the gall-bladder, and ' leaking ' duodenal ulcer were diagnoses offered in some of the 10 cases. Enteric fever, brucellosis, Weil's disease, and endocarditis were excluded when possible; amœbic abscess, hepatoma, perinephric abscess, and Charcot's intermittent hepatic fever were also considered.

Treatment and Mortality.—The treatment of multiple pyogenic liver abscesses must to-day still be surgical, for antibiotics alone, in such cases, will not cure the condition. This was seen in the patients presented here. Furthermore, without operation the diagnosis cannot be verified with safety. In the same way, cases of portal pyæmia in which only a clinical diagnosis is made, and in which a cure is claimed by the use of antibiotics alone, must be regarded with reserve as such cases are not verified.

The main difficulty of surgical treatment is to drain all the abscesses, for usually they lie deep within the liver, are many in number, and are separated one from the other. Sometimes, therefore, it will be impossible to drain them all. The approach best made is one that avoids contamination of pleural and peritoneal cavities. Often such an extra-serous approach is impracticable, as when at laparotomy the diagnosis is made by needling the liver. In such a case a gauze roll, left in situ for a while post-operatively, was found efficacious in packing the liver off from the general peritoneal cavity, thus preventing spread of infection.

If found, the primary focus may need obvious attention, but even if minor in degree it should be dealt with, as in this way the development of further abscesses from infected emboli is prevented. Venous ligation, e.g., of the ileo-colic or inferior mesenteric veins, is a further safeguard against emboli, but will be of little use if a suppurative thrombophlebitis extends into the portal vein itself.

The mortality *without drainage* was here 100 per cent, which is the same as it was in the days before antibiotics. Contributing to this figure were other complications of the primary focus of inflammation. Of the 2 cases drained, one died and in this patient a cerebral abscess had developed. Ochsner and others reported a mortality of 95 per cent for their 20 personal cases of multiple abscess, and contributing to this figure was the development of complications, either by direct spread of infection from the liver abscesses, or by emboli.

Since the introduction of antibiotics, cases of portal pyæmia, with recovery, have been reported—d'Abreu (1946); Flynn (1946); Hoffman, Partington, and Desanctis (1954)—and MacFadzean and others report a series of 14 cases of *solitary* pyogenic liver abscess with 100 per cent survival. In all these cases, some form of drainage had been established.

Just what effect antibiotics have had on the mortality of cases with multiple abscesses in which drainage has been performed must await the publication of a large series of such cases comparing the results with those of the pre-antibiotic era.

CONCLUSIONS

From consideration of 10 verified cases of portal pyæmia seen between 1951 and 1956, and from comparing these cases with what has previously been written in the literature on the condition, the following conclusions are drawn :—

1. The clinical picture to-day is unchanged from what it was in the pre-antibiotic era.
2. Diverticulitis coli, as the primary focus of infection, may contribute to a large percentage of the cryptogenic liver abscesses.
3. An obvious primary focus of inflammation is not necessary for a diagnosis of pyogenic liver abscess to be made.
4. Subphrenic abscess is not infrequently associated with liver abscess, and may distract the surgeon's attention from finding the liver abscess.
5. Antibiotics alone do not alter the course of the disease.
6. Drainage, together with antibiotics, offers the only hope of survival with multiple liver abscesses in portal pyæmia.
7. The primary focus should be dealt with to prevent further abscesses developing after drainage has been established.

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POST-OPERATIVE PSEUDOMEMBRANOUS ENTEROCOLITIS ASSOCIATED WITH STAPHYLOCOCCUS PYOGENES

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Many cases, fatal and non-fatal, of antibiotic-resistant staphylococcal enterocolitis have now been reported in this country (Kramer, 1948; Gardner, 1953; Hay and McKenzie, 1954; Williams, 1954; Frame and Short, 1955; Fowler, 1955; Barber and Burston, 1955; Todd and Hopps, 1955; and others). They include cases of post-operative staphylococcal enterocolitis where different antibiotics were used in the pre-operative preparation, and the *Staphylococcus pyogenes* isolated from the fæces or intestinal contents was found resistant to the corresponding antibiotic or antibiotics. These post-operative cases have followed extra-abdominal as well as abdominal operations. They also include cases of staphylococcal enterocolitis complicating the antibiotic treatment of medical diseases of different systems including the gastro-intestinal tract.

In contrast Williams and Pullan (1953) found no staphylococci in 6 of their 10 cases of pseudomembranous enteritis, but no pre-operative antibiotic therapy was used. Dawson-Edwards and Morrissey (1955) found staphylococci in only one of 8 cases of acute enterocolitis following partial gastrectomy; some had received antibiotics before operation, others only when the condition was established.

A leading article in the *Lancet* (1954), referring to post-operative pseudomembranous enterocolitis in patients who had not received antibiotics, states that staphylococci or other significant organisms had not so far been isolated from these cases. Barber and Burstol have since reported 2 cases of non-fatal post-operative enterocolitis with staphylococci in the fæces, and neither had received antibiotics before operation. One, and probably a further two of Williams and Pullan's cases belong to the same category.

A fatal post-gastrectomy case of pseudomembranous enterocolitis in a man who had not received antibiotic therapy, and from whose gastro-intestinal tract *Staph. pyogenes* was isolated during life, is recorded here.

CASE REPORT

The patient, a 50-year-old man, was admitted to hospital on Nov. 12, 1955, for a partial gastrectomy. He had complained of dyspepsia for six years with recent attacks of vomiting, and radiological examination showed a stenosing duodenal ulcer. His blood-pressure, hæmoglobin, and blood-cytology were normal. He received no pre-operative antibiotic or chemotherapy. A partial gastrectomy was performed in the early afternoon of Nov. 14. The stomach was aspirated by continuous suction through a Ryle's tube after the operation.

The patient's progress appeared satisfactory until the evening of the first post-operative day. His pulse rate was then found to be 110 per minute; he was restless, anxious, apyrexial, and complained of fatigue. Next morning his condition had deteriorated and he showed the picture of severe peripheral circulatory collapse. He was transfused with 2 litres of blood and a laparotomy was carried out for suspected internal hæmorrhage or leakage at the anastomosis. The anastomosis was healthy, and the peritoneal cavity and small intestine appeared normal.

During the next 24 hours he received 3 litres of normal saline, 2 litres of 5 per cent glucose in distilled water, and 1 litre of blood. His condition on the afternoon of the third post-operative day was still one of severe peripheral circulatory failure with a systolic blood-pressure of 60 mm. Hg. He now passed a liquid, offensive bowel motion, the first since his operation. Examination of his plasma showed urea 85 mg. per cent, protein 7·3 g. per cent, P.C.V. 65 per cent, chloride 85 mEq./l., sodium 140 mEq./l., potassium 5·6 mEq./l., bicarbonate 15 mEq./l. A presumptive diagnosis of pseudomembranous enterocolitis was made. Specimens of fæces, gastric aspirate, blood, and nasal swabs were collected for bacteriological examination. Erythromycin, noradrenaline, and 1 litre of fluid intravenously per hour were prescribed, but despite these measures the patient continued to deteriorate and developed fæcal incontinence. He died in the early hours of the morning of the fourth post-operative day, 14 hours after the presumptive diagnosis of enterocolitis was made. The patient received omnopon gr. $\frac{1}{3}$ and scopolamine gr. $\frac{1}{150}$ pre-operatively, and morphine gr. $\frac{1}{4}$ on the first and second post-operative nights.

Fig. 406.—A mucosal fold (valvula connivens) showing the pseudomembrane and complete mucosal necrosis of tip with preservation of the mucosal glands at sides and base. H. and E. (×12·5.)

Necropsy.—Necropsy was performed six hours after death, the body having been kept in the refrigerator during that time. The lungs showed basal congestion. The heart was normal; the coronary arteries showed a moderate amount of atheroma.

The abdominal cavity contained 1 litre of brownish fluid. The intestines were grossly distended with liquid fæces; the peritoneal surface was dull, cyanosed, and in places showed fibrinous peritonitis with fragile adhesions between the loops. The anastomosis between the stomach and jejunum was water-tight and healthy. The mucosa of the œsophagus, stomach, duodenum, and

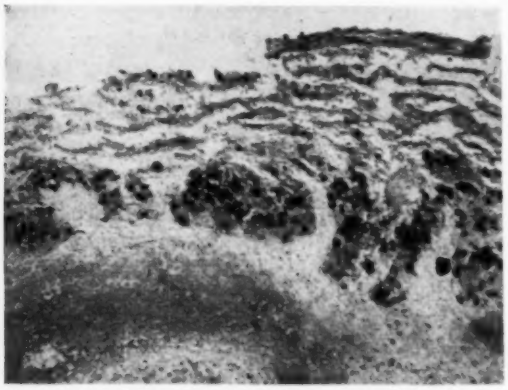

FIG. 407.—The pseudomembrane showing bacterial colonies confined to the luminal surface. Gram's stain. (× 80.)

the first 20 cm. of the jejunum showed no pseudomembrane or any evidence of inflammation. The mucosal surface of the remainder of the jejunum and the entire ileum were completely covered with a conspicuous yellowish-white friable pseudomembrane. It was noteworthy that there was not a square centimetre of the mucosal surface of the involved jejunum and the ileum which did not show the yellowish-white membrane. The mesenteric glands were inconspicuous The cæcum and ascending colon showed several large patches of membrane; the mucosal surface of the rest of the large bowel was reddish in colour but showed no membrane.

The liver was normal in size, and the cut surface was pale. The spleen was of normal weight, congested, and soft. The surface of normal-sized bisected kidneys showed pallor of the cortex.

HISTOLOGY.—Sections of the œsophagus, stomach, duodenum, and upper 20 cm. of jejunum were normal. Microscopical examination of the rest of the jejunum and the ileum showed necrotizing inflammation of the mucosa with pseudomembranous formation. The membrane was composed of necrotic cellular debris, fibrin, variable numbers of pus cells, and clusters of Gram-positive cocci and Gram-positive rods (*Fig.* 406). It was noteworthy that the bacterial colonies were largely confined to the luminal surface of the pseudomembrane (*Figs.* 407, 408). In the small intestine the mucosal necrosis varied from a superficial process where the basal parts of the glands were intact to one where the entire mucosa was necrotic. The tips of the mucosal folds (valvulæ conniventes) almost invariably showed complete mucosal necrosis with usually preservation of the muscularis mucosæ. The mucosa at the bases and between the folds was similarly involved in some areas, but in others only the superficial part was affected. The submucosa was œdematous; the submucosal venules and mucosal capillaries were widely dilated and occasionally showed fibrin thrombi. The muscular layers were not affected and the serosa showed deposition of fibrin. The histological features were essentially the same as those described by Penner and Bernheim (1939). In the ascending colon the pseudomembranous inflammation was confined to the superficial part of the mucosa with preservation of the bases of the tubular glands.

The liver showed areas of centrilobular degeneration and œdema; the spleen was congested; and the kidneys showed cloudy swelling of proximal convoluted tubules.

BACTERIOLOGY.—Aerobic and anaerobic blood-cultures were sterile after two weeks' incubation at 37° C. The nasal swabs cultured on blood-agar grew *Staphylococcus aureus*, *Streptococcus viridans*, and *Neisseria catarrhalis*.

Gram-stained films of the gastric aspirate and fæces collected during life showed a similar bacterial picture

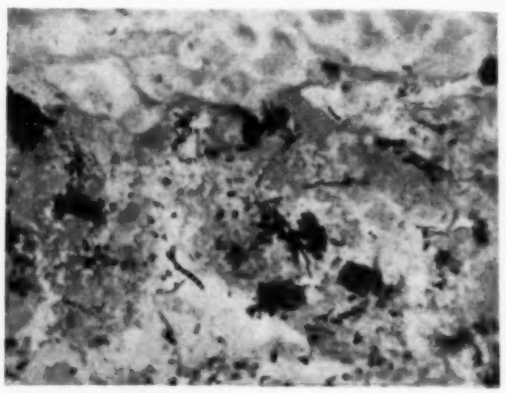

FIG. 408.—A higher power view of the luminal surface of the pseudomembrane showing colonies of staphylococci and Gram-positive rods. Gram's stain. (× 400.)

and consisted of encapsulated Gram-positive bacilli (*Clostridium welchii*), Gram-negative coliform bacilli, and Gram-positive cocci in pairs and small clusters.

Both the fæces and stomach aspirate were cultured on blood-agar aerobically and anaerobically, MacConkey's medium, nutrient agar containing 0·25 per cent beta phenylethyl alcohol (Lilley and Brewer, 1953), and in Robertson's cooked-meat medium. *Staph. aureus*, *Cl. welchii*, *B. coli*, and *Str. fæcalis* were isolated from both the fæces and the stomach aspirate.

The only additional information gained from bacteriological examination of post-mortem specimens was the isolation of a *Staph. aureus* from the liver, *Cl. welchii* from various levels of the gastro-intestinal tract, and the failure, presumably due to the erythromycin the patient received, to grow *Staph. aureus* from the alimentary tract despite its presence in sections.

The *Staph. aureus* isolated from the nose, gastric aspirate, and fæces during life and from the liver after death was coagulase-positive, sensitive to all the usual antibiotics, and belonged to the phage-pattern 52A/79. Professor C. L. Oakley (Oakley and Warrack, 1953) examined cultures of *Cl. welchii* for toxin production with the results shown in *Table I*.

The cultures are characteristic type A strains, and did not show heat-resistance.

DISCUSSION

The diagnosis of pseudomembranous enterocolitis was not made in this case until the patient had passed a diarrhœal motion and examination of the blood showed severe hæmoconcentration and metabolic hypochloræmic acidosis. Pettet, Baggenstoss, Dearing, and Judd (1954) remark that the explosive onset of watery diarrhœa has come to be associated with pseudomembranous enterocolitis in the minds of many clinicians. Of their 65 post-operative cases only about half were noted to have diarrhœa. On the other hand Williams and Pullan, and Dawson-Edwards and Morrissey found diarrhœa a prominent feature in practically all their cases following gastrectomy.

Antibiotic-resistant staphylococcal enterocolitis is at present thought to be due to infection by resistant

strains of staphylococci of the gastro-intestinal tract of patients debilitated by an operation or medical illness, and in whom the normal inhibitory intestinal flora has been suppressed or eliminated by one or more antibiotics. No antibiotics were administered in this case before the onset of the enterocolitis, and bacteriological examination of the fæces during life showed a normal fæcal flora. Indeed, in a post-operative case of pseudomembranous enteritis associated with a streptomycin-resistant staphylococcus, Corridan (1956) considered that the fæcal flora was unaltered. Apparently, then, some cases of enterocolitis associated with staphylococci do occur despite the presence of a normal fæcal flora.

Table I.—Result of Examination of Cl. Welchii for the Production of Toxins

Source of *Clostridium Welchii*	Toxins								
	α	β	δ	ε	θ	ι	κ	λ	μ
Stomach during life	tr.	–	–	–	+	–	+	–	–
Fæces during life	+	–	–	–	+	–	tr.	–	–
Small intestine P.M.	+	–	–	–	+	–	+	–	–
Small intestine P.M.	tr.	–	–	–	+	–	+	–	–
Colon P.M.	+	–	–	–	+	–	tr.	–	–

The sterilizing effect of the normal gastric juice is well known and the empty stomach is generally sterile. Cregan and Hayward (1953) found the jejunum and the greater part of the ileum without a resident flora and practically sterile. It is the opinion of Wilson and Miles (1955) that it is impossible at present to say how far this is attributable to the continuing effect of the gastric juice and how far to the separate antibacterial mechanism postulated by Cregan and Hayward.

Films and cultures of the stomach aspirate in this case during life showed that the bacterial content was similar to that found in the fæces. It appears likely, then, that the bacterial members of the fæcal flora invaded and colonized the small bowel and stomach from below. Since Brodie, Kerr, and Sommerville (1956) found that fæcal carriers of staphylococci are commonly also nasal carriers, it seems that staphylococci can reach the small intestine from the large bowel as well as from the nasopharynx, the more usually accepted site.

Because the gastric juice was continuously aspirated from the stomach remnant in this case, the amount reaching the small intestine must have been greatly reduced or negligible. In this respect it is well known that the administration of omnopon-scopolamine and morphine reduces the gastro-intestinal secretion and motility. It is noteworthy that enterotoxæmia of sheep, which is caused by epsilon toxin of *Cl. welchii*, type D, is difficult to produce experimentally. Successful attempts have been attributed to the administration of cultures along with belladonna and opium to paralyse intestinal movements (Bennetts, 1932). It appears then that the negligible amount of gastric juice entering the jejunum together with the administration of omnopon-scopolamine and morphine were possible factors in the failure of the antibacterial mechanism of the small bowel in this case.

Penner and Bernheim in a comprehensive study of acute post-operative enterocolitis found submucosal dilatation of capillaries and venules, followed by œdema of the submucosa and mucosa, and finally by focal mucosal necrosis. They consider that fusion and secondary infection give rise to the extensive pseudomembranous lesion. They think that the primary cause of these findings is shock due to the operation. Gesell (1919), as a result of his experimental findings, postulates that compensatory vaso-constriction resulting from shock may reduce the blood-flow below that necessary for normal nutrition and so cause mucosal necrosis and inflammation. The present case showed no overt evidence of shock during the operation or for twenty-four hours afterwards. Nevertheless, it is not impossible that the operation produced compensated shock and reduced the blood-flow to the intestinal mucosa below that sufficient for the effective operation of the antibacterial mechanism or normal nutrition.

The exact role of *Staph. pyogenes* in the pathogenesis of staphylococcal enterocolitis is still undecided, and some authors (Penner and Bernheim; Dearing and Heilman, 1953; Dawson-Edwards and Morrissey) ascribe only a secondary and accentuating one to it.

Howie, Duncan, and Mackie (1953) investigated the role, if any, of *Cl. welchii* of type A in the causation of diarrhœa following partial gastrectomy, and consider that the matter is still *sub judice* and needs re-examination. Although it colonized the stomach and small intestine during life, it is not possible to say whether it had any pathogenic significance or none in this case.

SUMMARY

Many cases of antibiotic-resistant staphylococcal enterocolitis have been reported in this country. They include non-operative as well as post-operative cases. In contrast, post-operative cases of pseudomembranous enterocolitis have also been reported in which *Staph. pyogenes* was not isolated.

A case of post-operative pseudomembranous enterocolitis in a man who had not received antibiotic therapy and from whose gastro-intestinal tract *Staph. pyogenes* was isolated during life is described.

The bacterial flora isolated during life from the gastric aspirate and fæces was similar and consisted of *Staph. pyogenes*, phage-pattern 52A/79, *Cl. welchii*, type A, *B. coli*, and *Str. fæcalis*. Nutrient agar containing 0·25 per cent beta phenylethyl alcohol was useful in the isolation of the staphylococcus in the presence of Gram-negative coliform organisms.

The small intestine showed necrotizing pseudomembranous inflammation with complete necrosis of the mucosa of the tips of the mucosal folds (valvulæ conniventes). The colonies of staphylococci were conspicuously confined to the luminal surface of the pseudomembrane. There were large patches of pseudomembranous colitis in the ascending colon.

It is suggested that interference with or inhibition of an antibacterial mechanism normally present in the small bowel may be concerned in its pathogenesis.

We are indebted to Professor C. L. Oakley for examining *Cl. welchii* for toxin production; and to Mr. N. Coward, Mr. R. G. Taylor, and Mr. A. Riley for technical assistance.

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VERTEBRAL OSTEOCLASTOMA WITH SPINAL CORD COMPRESSION*

BY NORMAN WHALLEY, SWANSEA

THE osteoclastoma has been known by many names in its time—myeloma, myeloid sarcoma, *tumeur à myéloplaxes*, benign giant-cell tumour, and even chronic hæmorrhagic osteomyelitis. Willis (1949) prefers the name 'osteoclastoma' because it correctly expresses the histogenesis of the tumour. Osteoblasts and osteoclasts are functional variants of cells of the same type. Osteoclastoma is a tumour of those cells in which the functional bias is osteoclastic rather than osteoblastic.

This paper is concerned with 4 examples of osteoclastoma of the vertebral bodies which resulted in pressure on the spinal cord. Three patients were treated personally and I have been able to follow their progress from 5 to 8 years. The fourth case was under the care of Professor Sir Geoffrey Jefferson and he has kindly supplied me with the clinical details. Primary vertebral neoplasms are uncommon, although from time to time such tumours as osteoma, chondroma, hæmangioma, sarcoma, Ewing's tumour, and chordoma may be encountered.

In recent years, attention has been drawn to the occurrence in long bones and the vertebral column of a lesion known as aneurysmal bone-cyst. Jaffe and Lichtenstein (1942), in a paper dealing with unicameral bone-cyst, pointed out that while the lining tissue of a bone-cyst might show giant cell reaction, there was no ground for postulating any relationship between bone-cyst and osteoclastoma. In reviewing their pathological material, they singled out as different and distinct 2 cases of a peculiar blood-containing, cyst-like lesion. The first of these occurred in the pubis of a youth aged 17 years. The second occurred in the body, neural arch, and spinous process of the second dorsal vertebra in a male aged 18 years. The expanded vertebral body was luxated anteriorly and there were signs of spinal cord compression. Following surgical exploration there remained a transverse myelitis. These distinctive cystic bone lesions later became known as aneurysmal bone-cysts when in 1950 Lichtenstein established that aneurysmal bone-cyst was indeed a separate pathological entity; he also pointed out how it could be confused with osteoclastoma. Reparative new bone formation is a feature of this lesion; it tends to heal spontaneously after incomplete removal or even without treatment at all. It is doubtful whether it can be included in any true tumour classification. Radiologically it would be easy to confuse this cystic lesion with osteoclastoma, but pathologically the two are quite unlike. Confusion in the past has arisen because of the presence of multinucleated giant cells around the areas of hæmorrhage in aneurysmal bone-cyst. These giant cells are smaller and have fewer nuclei than those of osteoclastoma; the fusiform or round-celled stroma so characteristic of osteoclastoma is not seen in aneurysmal bone-cyst.

Platt (1949) has stressed the need to differentiate between osteoclastoma and the dystrophic cysts of bone. Dystrophic cysts tend to spontaneous repair. The osteoclastoma, on the other hand, in its true guise, is always aggressive and destructive. The blood-chemistry has been normal in the 4 cases reported here. I have not seen any example of fibrocystic disease of bone which has given rise to spinal cord compression, but a case of vertebral involvement and paraplegia due to polyostotic fibrous dysplasia was recorded by Teng, Gross, and Newman in 1951. Pathologically, the affected area was cystic and contained giant cells with abundant fibrous and cartilaginous tissue.

In gross appearance an osteoclastoma tends to be uniformly red or dark red in colour. In parts it may be grey or of a fleshy consistency. In advanced untreated cases areas of hæmorrhage and necrosis may be seen. In long bones the affected end is expanded and the tumour is delimited by a very thin shell of bone. The microscopic appearance of osteoclastoma is characteristic (*Fig.* 409). The field

* Based on a paper read to the Society of British Neurological Surgeons at Södersjukhuset, Stockholm, on May 25, 1956.

consists of fusiform or rounded cells and multinucleated giant cells, together with the complete absence of any signs of osteoid or bony differentiation. The picture is unlike that of any other bony tumour.

The giant cells measure up to 100 μ or more in diameter and each possesses many nuclei. These nuclei are identical with those of the mononucleated cells and there are appearances which suggest that giant cells are formed by fusion of the mononucleated cells. Most osteoclastomas behave in a benign way, grow slowly, do not transgress the periosteum, and do not metastasize. They are curable by removal if this is possible. Incomplete removal is followed by recurrence. However, some tumours do transgress the periosteum and invade surrounding tissues and a few do metastasize to lymph-nodes and lungs. However, in the examples of spinal osteoclastoma previously recorded I have been unable to find a record of metastases.

Osteoclastomas are relatively uncommon tumours and probably their incidence is about half that of the malignant primary bone tumours. Kolodny (1927), in an analysis of the material of the Bone Registry, found that of all the osteoclastomas registered, 8 per cent occurred in the spine. At the Mayo Clinic in a series of 124 osteoclastomas only 2 vertebral tumours were found (Myerding, 1941). Coley (1949) in his book, *Neoplasms of Bone and Related Conditions*, listed 509 cases of osteoclastoma of which only 3 were in the spine. In 1949 three separate papers were published in Britain by Ellis, Prossor, and Windeyer and Woodyatt in which they reviewed the treatment of osteoclastomas in the bones of the skeleton. There were 84 cases of osteoclastoma studied in all and in 11 cases the tumour was in a vertebra or the sacrum. Only sparse details are available with regard to 5 of these 11 cases. One of them was not verified histologically; 1 died with a cauda equina lesion; 2 sacral cases developed fistulæ; and the fifth case developed a cerebrospinal fluid leak after implantation of radon seeds.

I have been able to trace 33 cases of spinal osteoclastoma in the literature—cases which have been studied in any detail. Of these, 18 had evidence of spinal cord or cauda equina involvement; 1 resulted in a brachial plexus syndrome and another in a sciatic syndrome. In all but one case an attempt at pathological diagnosis seems to have been made. At least 5 of these patients died, 1 after four years. Five patients were well four, seven, seven, ten, and twenty-two years after treatment. The follow-up periods of most of the remaining cases at the time of publication were short. *Table I* gives some details of cases previously recorded as examples of osteoclastoma of the spinal column. It may well be that some of them, if reviewed in the light of more recent knowledge, would be included in the category of aneurysmal bone-cyst. This is particularly true in those instances where the lesion involved the spinous process or lamina, as these sites seem to be more frequently involved in aneurysmal bone-cyst rather than the vertebral bodies. Besse, Dahlin, Bruwer, Svien, and Ghormley (1953), Barnes (1956), and Taylor (1956) all mention the likelihood of cord involvement in aneurysmal bone-cyst, but they do not mention any particular instances of such occurrence.

CASE REPORTS

Case 1.—A young girl of 12 years whom I saw for the first time in September, 1948. For four weeks she had complained of weakness of both legs. This had progressed rapidly. She had paræsthesiæ in both legs and girdle pains beneath the right and left costal margins. There was no sphincter disturbance. On examination there was a kyphoscoliosis in the lower dorsal region of the spine; she was unable to walk or stand because of weakness of the lower limbs. There was diminution of all forms of sensation below the umbilicus. There were pyramidal signs in both legs.

Lumbar puncture and Queckenstedt's test showed complete spinal block with xanthochromic cerebrospinal fluid.

Radiographs of the spine showed a tumour arising in the left side of the body of the tenth dorsal vertebra,

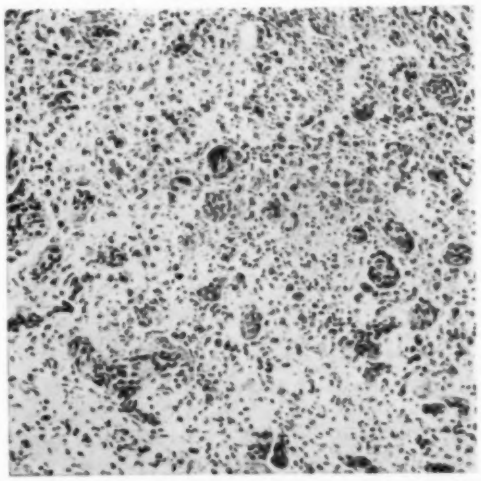


FIG. 409.—The field shows fusiform and rounded cells with many multinucleated giant cells. There are several very large giant cells to the left of the field. There is no sign of osteoid or bony differentiation.

involving the pedicle, transverse process, and half the lamina. There was trabeculation to be seen in this area (*Fig.* 410). In addition there was partial collapse to the left side and anteriorly. The pre-operative diagnosis was osteoclastoma.

At operation on Sept. 29, 1948 a laminectomy approach was used. A mass of tumour was uncovered in the region of the interarticular joint on the left side. This had a very thin shell of bone. The laminectomy was extended upwards and downwards revealing a mass of extradural tumour which was encircling and constricting the spinal dura mater and cord. The extent of tumour spread within the spinal canal may be seen in the radiograph (*Fig.* 410) which shows the extent of the laminectomy. The tumour was red in colour and vascular. As much as possible was removed.

Following operation the patient was nursed on a plaster-of-Paris bed. There was rapid return of motor and sensory function. She was given a course of radiotherapy—2000 r in fourteen treatments. This was followed by another course of 1800 r three months later. She remained on the bed for five months.

The histological report showed that the tumour was an osteoclastoma.

Present radiographs show that recalcification has occurred (*Fig.* 411). Eight years after operation the patient is extremely well and there are no symptoms. There is a residual kyphoscoliosis. There are no abnormal neurological signs. She was married a few months ago.

Table I.—DETAILS OF RECORDED CASES OF OSTEOCLASTOMA OF THE SPINAL COLUMN

No.	Author	Sex	Age	Year	Site	Symptoms	Signs	Biopsy-Histology	Treatment	Result
1	Bowman and Reals	F.	18	1955	Destructive process in spinous process of 6th cervical vertebra with pathological fracture	Pain in neck	Nil	Benign giant-cell tumour with pathological fracture	Deep X-ray therapy	Healing
2	Brock and Bogart	M.	20	1945	Destructive lesion involving spinous process, lamina, inferior facets and portions of superior facets, 4th cervical vertebra	Pain in right arm	Nil	Giant-cell tumour	Deep X-ray therapy	Recalcification
3	Hamsa and Campbell	F.	6	1953	Destructive lesion right half first sacral vertebra going into adjacent wing of ilium	Pain in back and left hip (fall downstairs)	Swelling left side lower back	Benign giant-cell tumour	Deep X-ray therapy	Not stated
4		F.	14	1953	Destructive lesion left half 12th dorsal vertebra	Pain in low back after direct blow	Nil	Giant-cell tumour	Deep X-ray therapy and spinal fusion	Well after 10 yr.
5	Kaplan	F.	22	1942	Lesion in lamina of 2nd dorsal vertebra	Spastic paraplegia (8 days)	Spastic paraplegia	Giant-cell tumour	Laminectomy and removal	Complete recovery
6	Teng, Gross, and Newman	M.	22	1951	Lesion in lamina of 3rd lumbar vertebra	Weak left leg	Incomplete cauda equina lesion	Giant-cell tumour	Laminectomy and deep X-ray therapy	Recovery at time of report
7		F.	15	1951	Lesion in 6th cervical vertebra	Quadriparesis	Quadriparesis	Benign giant-cell tumour	Laminectomy, plaster-of-Paris cast, deep X-ray therapy	Improvement (no dates)
8	Lewis	F.	7	1924	Rarefaction right half body of 4th dorsal vertebra	Rapid onset of partial transverse lesion spinal cord	Paraparesis	Giant-cell tumour	Decompression, operation and radium 1 mth. later	Well recovered after 2 yr. 9 mth.
9	Madelung*	M.	19	1909	Cervical region	Motor and sensory disturbances	Paraparesis	Giant-cell myeloid sarcoma	Curettage	Well 3 mth. later
10	Schumann*	M.	27	1898	Lumbar region	Pain in back; tiredness	Paraplegia	Diagnosis—giant-cell sarcoma	Incomplete removal with recurrence	No result given
11	Manheimer*	F.	16	1911	Transverse process 2nd lumbar vertebra	Pain radiating to both legs	Not stated	Osteoid spindle-cell sarcoma with myxomatous areas and giant cells	Removal as far as possible	Well 5 mth. later
12	Sick*	M.	Not stated		Mass lumbar region between iliac crests and 12th rib	Not stated	Not stated	Giant- and mixed-cell fibrosarcoma	Partial removal Potassium arsenite	4 yr. later well
13	Ashurst*	M.	14	1913	Fourth and 5th lumbar vertebræ	Mass in right loin	Not stated	Giant-cell sarcoma	Operation and Coley's fluid	Well 7 yr. after
14	Harmer*	M.	16	1915	Lumbar region spinous process 3rd lumbar vertebra	Injury 3 mth. before. Pain and tenderness spine	Paraparesis	Giant-cell sarcoma	Removal and Coley's fluid	Well 2 yr. after
15	Rasch*	Not stated		1922	Tenth and 11th dorsal vertebræ	Not stated	Paraplegia	Giant-cell tumour (sarcoma)	Not stated	No result given
16	Mixter*	M.	31	1922	Fourth and 5th dorsal vertebræ	Girdle pains, weakness, numbness of legs, paralysis	Paraplegia	Not stated	Incomplete removal and Coley's fluid. Rœntgen ray	Recovering, but no result given
17	Coley*	M.	21	No date given	Not stated	Not stated	No paraplegia	Diagnosis of atypical giant-cell tumour	Coley's fluid	Well 22 yr. later
No.	Author	Sex	Age	Year	Site	Symptoms	Signs	Biopsy-Histology	Treatment	Result
18	Ewing	M.	20	1928	Lumbar region	Not stated	Not stated	? Giant-cell	Removal	Result not known

No.	Author	Sex	Age	Year	Site	Symptoms	Signs	Biopsy-Histology	Treatment	Result
17	Coley*	M.	21	No date given	Not stated	Not stated	No paraplegia	Diagnosis of atypical giant-cell tumour	Coley's fluid	Well 22 yr. later
18	Follis*	M.	40	1920	Mass in dorsal region near angle of left scapula	Not stated	Not stated	? Giant-cell tumour ? Sarcoma	Removal	Result not known
19	Cushing*	F.	13	1922	Dorsal region	Paralysis. Loss of sensation both legs	Paraplegia	Giant-cell tumour	Operation (laminectomy)	No recovery
20		M.	16	No date given	Lumbar region	Mass in back, pain lower dorsal and lumbar region	Paraparesis	Giant-cell sarcoma	Partial removal, deep X-ray therapy	Well 7 yr. later
21	Madelung*	M.	25	1890	Lumbar region	Not stated	Not stated	" Osteoid sarcoma with giant cells "	Partial removal	Did well for 4 yr. Two more operations. Died
22	Thompson*	M.	24	No date given	Sacrum	Not stated	Cauda equina lesion	" Giant tumour of benign type "	Operation given up because of hæmorrhage	Died after 4 mth.
23	Leake*	M.	Adult	1923	Lumbar vertebræ 1 and 2	Not stated	Cauda equina syndrome	Benign giant-cell tumour	No operation	Died
24	Cushing*	F.	13	1915	Sacrum	Pain lumbar region; incontinence of urine, no apparent paralysis	Not stated	" Sarcoma " of sacral canal with giant cells	Partial removal	Death after 4 yr. 4 mth.
25	MacFarlane and Linell	M.	35	1934	Body of 3rd cervical vertebra	Sudden pain in neck while cranking car. Crush fracture 3rd cervical vertebra	No paraplegia	Giant-cell tumour	Head extension on Bradford frame	Recovery for 9 mth. Working. Died of head injury
26	Ratko	F.	19	1955	Punched out defect 6th cervical vertebra. 'Washed out' 5th and 7th cervical vertebræ. Absence of intervertebral disks between them	Pain, brachial plexus syndrome left arm	Lump left side neck; dysphagia	Giant-cell tumour	Deep X-ray therapy and cervical collar	Well 2½ yr. after first seen
27	Santos	M.	27	1930	Gross destruction body 4th lumbar vertebra	Pain left groin and back; incontinence of urine; weakness legs	Conus syndrome	Benign giant-cell tumour	Laminectomy, deep X-ray therapy	1 yr. after, still retention of urine. No anæsthesia
28	Rogers†	M.	27	1903	Lumbar region	Not stated	Paraplegia	Giant-cell sarcoma	Biopsy, and Coley's fluid	Recovered to walk
29	Cumarati†	M.	25	1927	Atlas and odontoid. Forward subluxation of atlas	Not stated	Tetraplegia; bulbar palsy	Giant-cell tumour		Death
30	Adson†	M.	14	1928	Destruction right half 10th dorsal vertebra	Not stated	Paraplegia	Benign giant-cell tumour	Laminectomy	Complete recovery 2 yr. 9 mth. later
31		F.	13	1928	Lamina 2nd and 3rd cervical vertebræ	Pain in neck; mass	No paraplegia	Giant-cell tumour	Removal	Well 5 mth. after
32	Cotton†	M.	15	1928	Left side body, 5th lumbar vertebra	Fall on buttocks. Pain in back. Kyphosis; 4th and 5th lumbar vertebræ; mass fluctuant	Partial cauda equina lesion	Giant-cell tumour	Exploration and deep X-ray therapy	Well 2 yr. after. Mass disappeared
33	Verbeist	F.	35	1947	Transverse process 5th lumbar vertebra	Sciatica	No paralysis	*Tumeur à myéloplaxes*	Curettage and grafting later	Recovery

* Quoted by Lewis, D. (1924). † Quoted by Santos, J. V. (1930).

Case 2.—A young female aged 15 years when first seen. For six months she had complained of pain in the dorsal region of her spine associated with gradually increasing weakness of the lower limbs. I saw her in October, 1949, and at that time there was gross weakness of the legs with spasticity and pyramidal signs. She was unable to stand or walk. There was no sphincter disturbance. There was diminution of all forms of sensation below the umbilicus.

FIG. 410.—Radiograph (*Case* 1) showing tumour arising in left half of body of 10th dorsal vertebra. The tumour also involves the pedicle, transverse process, and half the lamina. Trabeculation is well marked. The amount of tumour removed from the extradural plane may be judged from the extent of the laminectomy.

Radiographs of the spine showed a tumour arising in the left half of the body of the tenth dorsal vertebra. This tumour extended into the adjacent pedicle and transverse process. Again there was an appearance of rarefaction and trabeculation. The appearance suggested that it was an osteoclastoma (*Fig.* 412).

Lumbar puncture and manometric tests again revealed complete spinal block with xanthochromia. Myodil injected into the cisterna magna was arrested opposite the body of the ninth dorsal vertebra (*Fig.* 413).

Operation exposed a tumour covered with a very thin shell of bone. As the laminectomy was continued a mass of red tumour was uncovered encircling and constricting the spinal dura mater and cord. The upper and lower limits of spread were identified and as much tumour as possible was removed. At the end of the operation a cavity remained in the body of the tenth dorsal vertebra.

The patient was nursed on a plaster-of-Paris bed, and post-operatively there was rapid return of motor and sensory function. She remained on the bed for five months and had two courses of deep radiation therapy. The first course consisted of 2000 r in fourteen treatments; the second course was given three months later and consisted of a further 2000 r in fourteen treatments.

The histological report showed that this was an osteoclastoma.

It is now almost seven years since she was first seen and operated upon. She is well and working regularly as a hairdresser. She can walk many miles; she swim and goes to dances. There is occasional urgency o micturition. On examination there are no residu abnormal neurological signs.

Radiographs (*Fig.* 414) show the presence of cavity in the body of the tenth dorsal vertebra. Th

FIG. 411.—Recent radiograph (*Case* 1) to show recalcification and healing following deep radiation therapy.

margins of the cavity are sclerosed, but the sclerosis i not so marked as in the previous case.

Case 3.—A female of 29 years. She was seen for th first time in October, 1951. For nine months she ha been having severe pain in the right hip. This wa associated with pain beneath the right costal margin. I addition she had been having pain in the lower dorsa region of the spine. During the six weeks before he admission to hospital she had complained of gradually increasing weakness of the left leg. There was n sensory or sphincteric disturbance.

On examination she was unable to stand or walk because of severe pain in the back and weakness of th left leg. There were pyramidal signs in the left leg and there was a marked kyphosis at the level of the twelfth dorsal spinous process. There was local tenderness a this site.

Radiographs (*Fig.* 415) of the spine showed gross collapse and wedging of the body of the twelfth dorsa vertebra; posteriorly there was obvious rarefaction and suggestion of trabeculation extending into the pedicl (*Fig.* 415 B). This could well have been a metastasis, o course, although there was no primary neoplasm t be found. It seemed possible that this was a osteoclastoma. There was a history of suspected pulmo nary tuberculosis in childhood, and the possibility o tuberculous osteitis had to be considered in the differenti diagnosis although the lung fields were clear on radi ology. Another anteroposterior radiograph showed soft-tissue shadow around the area of collapse whic could have been tumour or possibly a cold absces (*Fig.* 416).

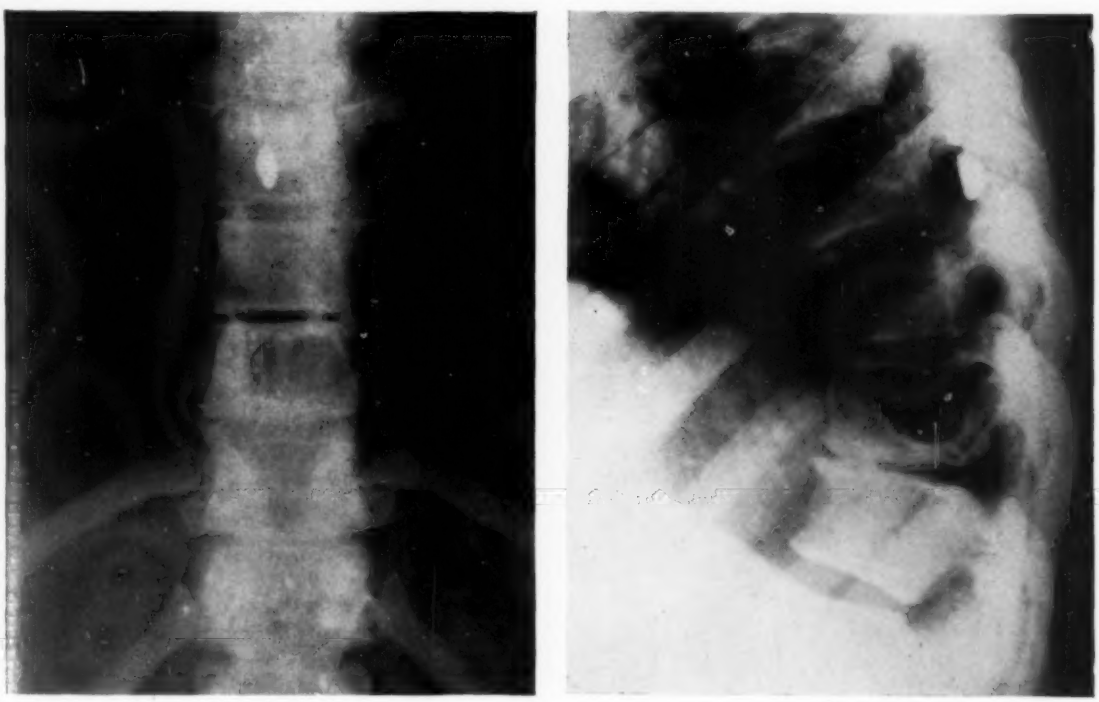
I felt that if the diagnosis of osteoclastoma could b established it would be worth while treating this patien with radiotherapy alone, as there was no spinal block o

manometric tests and she was by no means as paralysed as the first two patients.

After removal of the left twelfth rib the tumour was inspected. It was smooth and soft. There was a thick capsule but no bony shell. Attempted aspiration was negative, and after opening the capsule several large pieces of soft reddish-grey tumour were taken for section. Post-operatively the patient was nursed on a plaster-of-Paris

FIG. 412.—Anteroposterior radiograph (*Case* 2) to show the tumour arising in the left half of body of the 10th dorsal vertebra. The tumour extends into the transverse process. Myodil is arrested opposite the body of the 9th dorsal vertebra.

FIG. 413.—Lateral radiograph (*Case* 2) showing tumour in body of 10th dorsal vertebra extending into pedicle. Myodil is arrested opposite the body of the 9th dorsal vertebra.



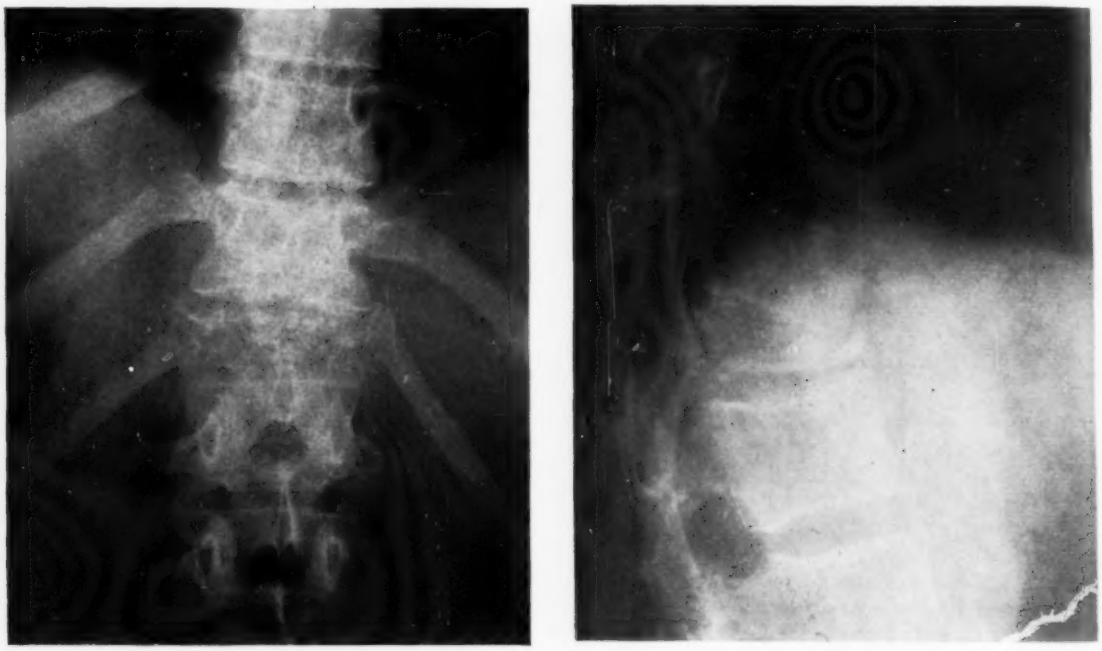
A B

FIG. 414.—A, Anteroposterior, and B, lateral radiographs (*Case* 2) to show appearance almost seven years after operation and radiotherapy. There is a cavity in the body of the 10th dorsal vertebra with sclerosed edges.

bed. Histological examination showed that this was an osteoclastoma.

In November, 1951, she was given a course of radiotherapy, 2400 r in three weeks. Following this she made a rapid recovery; the pain disappeared and the paresis of the left leg recovered. She is well at the present time. There is no pain. There is good spinal movement. There is no weakness of the legs and there are no residual abnormal neurological signs. The patient is working for eight hours a day behind the counter of the local post office. Recent radiographs show marked evidence of recalcification, although there is a severe degree of kyphosis (*Fig.* 417).

Case 4.—A young male, aged 16 years. He came under the care of Professor Sir Geoffrey Jefferson in August, 1939. At that time he was completely paraplegic and lying on a plaster bed. Several months before he had been hit on the back by a fellow midshipman and almost immediately he had become weak in the legs. The weakness progressed until finally he was completely paraplegic with a sensory level corresponding to the fourth thoracic segment. It was impossible to palpate the second thoracic spinous process, which appeared to have been replaced by an area of induration in the overlying skin. There was marked tenderness at this site. Radiographs of the spine showed absence of the second thoracic spinous process, together with absence of the lamina of the second thoracic vertebra. The pedicles could be seen on both sides and appeared to be normal. On two of the radiographs a very faint soft-tissue shadow could be seen, suggesting that the lesion was either an abscess or a tumour. It was felt that the most likely cause of the paraplegia was a bony neoplasm, since tuberculosis is such a rare condition at this site. At operation, a tumour involving the spinous process and lamina of the second thoracic vertebra was explored and removed. The tumour had been compressing the spinal cord. At the end of the operation the spinal compression was completely relieved. Following the tumour removal, treatment was continued on the plaster-of-Paris bed and a course of deep X-ray therapy was given. There was gradual recovery of function in the legs and six months after operation the patient was able to walk by himself without support. Eleven months after operation he was leading a normal life and indulging in the ordinary physical activities for a boy of his age. All told he had 6000 r units of deep X-ray therapy. This patient has been seen at regular intervals since his operation in 1939. He remains well and there are no residual abnormal neurological signs. The bony tumour was examined by Professor S. L. Baker, who diagnosed it as an osteoclastoma.



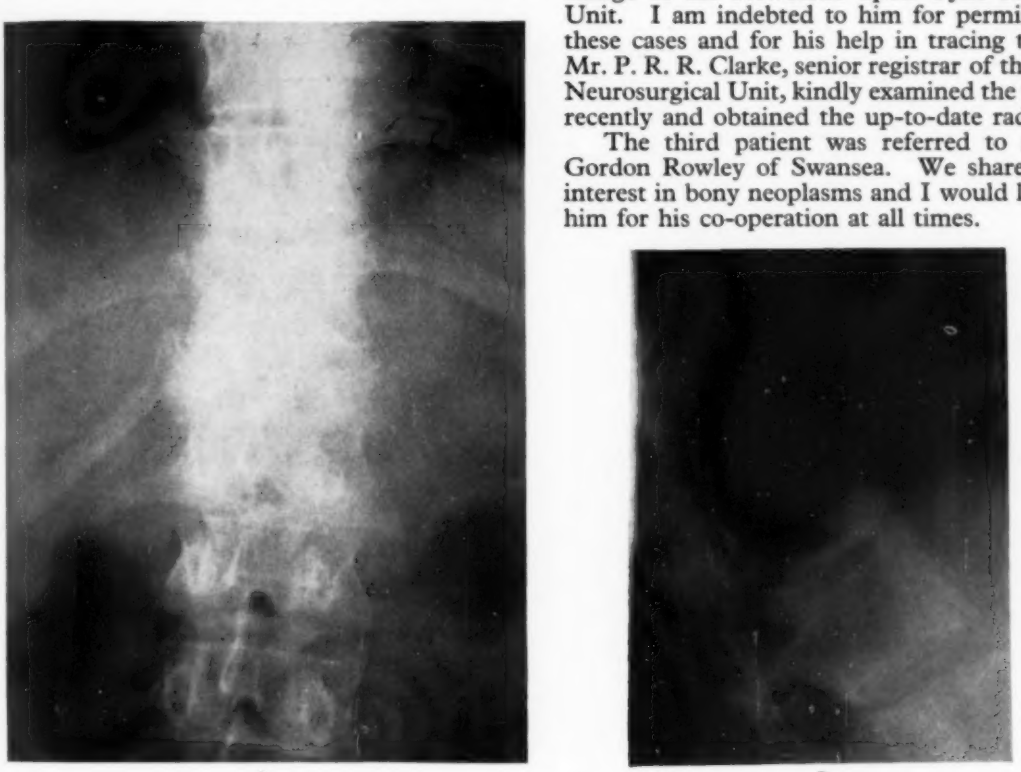
A B

FIG. 415.—A, Anteroposterior, and B, lateral radiographs (*Case* 3) showing gross collapse and wedging of body of the 12th dorsal vertebra. Posteriorly there is obvious rarefaction which extends into the pedicle.

FIG. 416.—Anteroposterior radiograph (*Case* 3) showing soft-tissue shadow around area of collapse. This appearance could have been due to tumour or possibly an abscess.

CONCLUSIONS

Four cases of vertebral osteoclastoma with spinal cord compression have been described. Each case has been verified histologically. They are unusual lesions and very few cases have been described in the literature. All four cases made good recoveries; three of the cases were treated by surgical decompression of the spinal cord supplemented with deep radiation therapy; the other case was treated by radiotherapy alone once the diagnosis had been established histologically. Previously recorded instances of spinal osteoclastoma have been reviewed; it is suggested that some of these cases may fall into the category of aneurysmal bone-cyst.



A B

FIG. 417.—A, Anteroposterior, and B, lateral radiographs (*Case* 3) taken almost five years after the course of radiotherapy. There is marked evidence of recalcification in the collapsed area.

There can be no question that radiotherapy alone can produce remarkable results in osteoclastoma of the long bones. Recalcification usually occurs gradually and progressively over a period of from one to two years. Most authors stress the need for biopsy and accurate verification before treatment is begun, although this is not a universal practice. Lichtenstein (1952) points out that the diagnosis of osteoclastoma cannot always be made on the radiographic appearances alone and that other non-tumorous and tumorous lesions may resemble it. He says that recognition of an osteoclastoma in sites other than the long bones is often a difficult problem. It seems clear that microscopic examination of the lesion is the only certain way to be sure that one is dealing with an osteoclastoma or not. *Case* 3 did well with radiotherapy alone after the nature of the lesion had been verified by histological examination. It seemed reasonable to do this as there was not complete paralysis. However, when paralysis is dense and spinal block is absolute (as in *Cases* 1, 2, and 4) one would be very bold indeed to pin one's faith on radiation alone without first relieving the cord compression surgically.

The first two patients were treated by me when I was deputy to Mr. G. F. Rowbotham, surgeon in charge of the Newcastle upon Tyne Neurosurgical Unit. I am indebted to him for permission to use these cases and for his help in tracing the patients. Mr. P. R. R. Clarke, senior registrar of the Newcastle Neurosurgical Unit, kindly examined the two patients recently and obtained the up-to-date radiographs.

The third patient was referred to me by Mr. Gordon Rowley of Swansea. We share a common interest in bony neoplasms and I would like to thank him for his co-operation at all times.

The fourth patient was under the care of Professor Sir Geoffrey Jefferson and has been examined regularly by him since 1939. I wish to thank him for permission to publish the case details.

The pathological examinations of *Cases* 1 and 2 were made by Dr. Ian Rannie and Dr. R. Schade, of the University Department of Pathology, Newcastle upon Tyne. Dr. Mary Sharp, of Swansea, examined the material from *Case* 3. The pathological examination of the tumour removed from *Case* 4 was made by Professor S. L. Baker, Professor of Pathology in the University of Manchester. I thank them all for their help.

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ABERRANT MUCOSA IN THE VERMIFORM APPENDIX

BY D. J. B. ASHLEY

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ABERRANT epithelial tissues have been described in many situations in the body, but apparently in no instance has ectopic mucosa been described in the appendix. The following case is presented because of the obvious rarity of such an event in this organ.

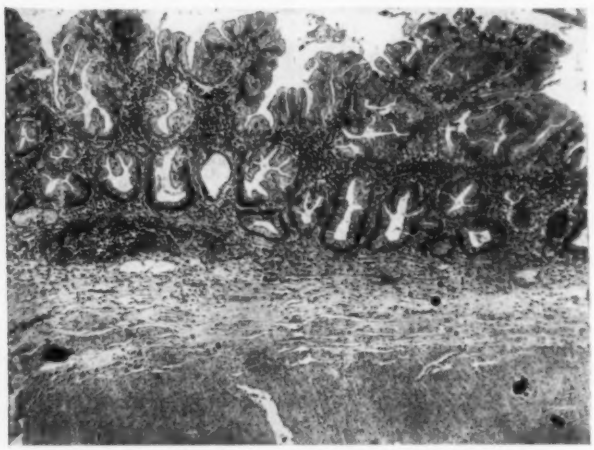

FIG. 418.—The lining of the appendix showing tall columnar epithelium overlying a glandular mucosa. H. and E. (× 60.)

CASE REPORT

The patient, a married woman 26 years of age, was admitted to this hospital in November, 1955, eleven weeks after the birth of her first baby, complaining of abdominal pain which had been present for only a few days. There was no relevant personal or family history and the diagnosis of subacute appendicitis was made. The symptoms subsided under medical treatment and she was discharged from hospital. Five months later she was re-admitted for interval appendicectomy which was carried out in April, 1956. At operation the terminal ileum, mesentery, and the right ovary were noted to be normal; there was no Meckel's diverticulum and no staining of the peritoneum suggestive of previous hæmorrhage. The post-operative course was uneventful and the patient is now well.

PATHOLOGICAL EXAMINATION.—The specimen removed was an appendix approximately 10 cm. long which showed no macroscopic abnormality on the serosal or the mucosal surface. Histological examination showed no abnormality in the peritoneum or in the muscularis propria, which invested the whole appendix in the normal manner. The proximal end was lined by normal appendicular mucosa which showed lymphoid hyperplasia and a diffuse infiltration with plasma cells and lymphocytes; occasional eosinophils also were present; the lumen contained fæcal material and large numbers of mononuclear cells. The distal 2 cm. of the appendix was lined by an entirely different type of mucosa. In this situation there was an inner lining of large palely eosinophilic columnar cells which contained no vacuoles and were arranged in a villous pattern with some pseudostratification; beneath this lining were gland acini which were formed of goblet cells and large pale cells; the glands were larger than those in the appendicular mucosa and frequently showed a wavy outline. This part of the mucosa contained a very few lymph-follicles, the mucosal infiltration was less marked, and the resemblance to the mucosa of the small intestine (*Figs.* 418–420) was most striking.

FIG. 419.—Showing detail of the epithelium lining the appendix and the glands containing goblet cells. H. and E. (× 88.)

DISCUSSION

An atypical mucosal lining to the appendix is apparently an excessively rare occurrence, a curiosity —this despite the many thousands of appendices which are examined in the histological laboratories of the world every year.

Epithelium, atypical to the organ in which it is found, is a relatively common finding on microscopical examination and is considered to be of several types. Metaplasia, a state in which the epithelium

consists of cells different in type from those normally found in the part, is commonly seen in states of chronic inflammation or of irritation. The change observed is usually from columnar to squamous epithelium and is well seen in the cavities of a bronchiectatic lung where the normal ciliated columnar cells of the bronchial mucosa are replaced by stratified squamous epithelium. Less common causes of this type of change are vitamin A deficiency and primary neoplasia (Anderson, 1948). This is a cytological rather than a histological change and is by far the commonest form of metaplasia. Much less commonly an abnormal tissue is found which is regarded as metaplastic. Arey and Bothe (1950) found that a high proportion of their cases of chronic gastritis showed, in the stomach, islands of mucosa of intestinal type; they did not find this type of mucosa in the stomach of a fœtus or in an adult stomach which showed no evidence of inflammation, and they regarded such a change as an atypical regeneration in response to irritation. A similar hypothesis has been postulated to account for the islands of intestinal epithelium sometimes seen in the gall-bladder mucosa in chronic cholecystitis, cholecystitis glandularis proliferans (King and MacCallum, 1931). The appearance in the present case could conceivably be regarded as analogous to those of Arey and Bothe and of King and MacCallum, but the great rarity of the condition compared with the high frequency of inflammatory disease in the appendix suggests that this may not be the correct explanation.

Nicholson (1950), who was of the opinion that normally epithelial differentiation is held in check by the environmental conditions, postulated a second form of epithelial atypia which he called prosoplasia; This is a state in which the epithelium undergoes differentiation in excess of that normal for the part, and a typical example of this type of change is in the urinary tract. Nicholson regarded the transitional epithelium of the urinary tract as an incompletely differentiated squamous epithelium and suggested that after changes in the environment full differentiation can take place. In the case reported above this explanation is excluded, as both appendicular and intestinal mucosæ are tissues fully differentiated along different lines and are not two stages in the same process of differentiation.

Structures may be present in abnormal situations as a result of aberrant differentiation and evocation. This is a not uncommon occurrence, and although the outstanding example is the presence of gastric mucosa in Meckel's diverticulum, many others may be seen: for example, pancreatic tissue, including islets of Langerhans, in the wall of the gut, islands of gastric mucosa in the lower part of the œsophagus, and a wide variety of mucosæ and glandular structures in the walls of alimentary reduplications and of intrathoracic developmental cysts. The author regards this as the explanation of the presence of mucosa of the small intestine type in this appendix.

The appearance of the mucosa at the distal end of this appendix must be distinguished from that seen in mucocele of the appendix. Woodruff and McDonald (1940) divided this condition into two groups: the first, which bears no relation to the present case, is associated with an obstruction at the base of the appendix which is lined by a thin mucosa consisting largely of mucous secreting cells; the second group, which they found to consist of only 10 cases in a review of 43,000 appendicectomies, they regarded as Grade I cystadenocarcinoma of the mucous lining of the appendix. It is this type of mucocele which is liable to cause pseudomyxoma peritonei. The atypical mucosa in the present case

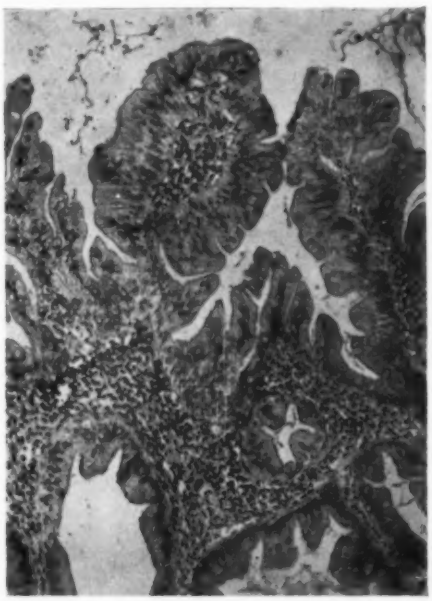

FIG. 420.—Showing the villous processes clothed by tall pale columnar cells which show pseudostratification. H. and E. (× 135.)

showed no evidence of neoplasia of this type; the cells were well differentiated, there was a high degree of tissue differentiation, and there was no evidence of distension of the appendix by the excessive production of mucus.

SUMMARY

A case of appendicitis is described in which the appendix contained an island of mucosa of small intestinal type. This is probably the first such case to be reported. It is considered that the aberrant mucosa is the result of a developmental anomaly of differentiation and not an example of tissue misplaced during ontogeny.

I am indebted to Mr. J. B. Oldham, under whose care the patient was admitted to hospital, for permission to publish this case.

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CHOLECYSTITIS CYSTICA AND RELATED LESIONS

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THE presence of minute mucosal pouches within the wall of the gall-bladder was described by Rokitansky (1842) and by Luschka (1858), although it was Aschoff (1905) who first appreciated their importance in betokening the previous occurrence of cholecystitis. These sequestrated mucosal acini became known as Rokitansky–Aschoff (R–A) sinuses. Later authors (e.g., Moore, 1937; Robertson and Ferguson, 1945) have emphasized the frequent association of R–A sinuses with cholelithiasis, the majority attributing their production to the effects of increased intracystic pressure. Some have, however, been more impressed by the proliferative nature of the lesions accompanying the presence of gall-stones and have discounted the importance of inflammation. Terms such as "adenomyoma" (Shepard, Walters, and Dockerty, 1942), "adenoma" (Mayo Robson, 1905) have been introduced to describe a condition, also known as "cholecystitis glandularis proliferans cystica" (King and McCallum, 1931; King, 1953), which affects an area at the apex of the gall-bladder in most instances. Other abnormalities, such as diverticula and the phrygian cap deformity (Bartel, 1916) which also occur mainly at the fundus of the viscus, have been described as separate entities.

When R–A sinuses occur, they tend to be most numerous at the fundus, where they may become large enough to be seen macroscopically. In such cases when palpating the gall-bladder at operation prior to removal, a nodule can be felt at the fundus which in most instances is found to contain a small diverticulum. Histologically the fundal area in these cases shows the changes described by King. Small pouches have also been observed radiologically in cholecystograms (March, 1948; Lawler, 1952; Sukman and Daron, 1953); their presence has been taken as evidence of the existence of gall-bladder disease.

Table I.—CLINICAL AND PATHOLOGICAL DATA IN 16 CHOLECYSTECTOMIES

Case number	1	2	3	4	5	6	7	8	9	10	11	12	13	14	15	16
Age	53	63	67	61	67	66	47	28	52	64	43	74	49	39	57	63
Sex	M.	F.	F.	F.	F.	F.	F.	F.	F.	M.	M.	M.	M.	M.	F.	F.
History	12 yr.		3 yr.	3 yr.	2 yr.	3 mth.	2 yr.	18 mth.	6 mth.	1½ yr.	3 yr.	6 yr.	3 mth.	6 yr.	5 yr.	2 mth.
Gall-bladder: dilated	+	+	+	+		+	+	+	+	+	+	+	+	+		+
normal					+										+	
contracted																
Wall: thin							+									
normal		+	+	+	+	+		+	+	+	+	+	+	+	+	+
thick																
Bile stained			+	+	+	+	+	+	+			+	+	+		
Stones					+			+	+	+	+		+	+	+	+
Fundus: diverticulum			+	+												
thickening	+	+	+	+	+	+	+	+	+	+	+	+	+	+	+	+
R–A sinuses					+			+	+	+						
Fibrosis			+	+	+	+	+	+	+	+	+	+	+		+	+
Mucosal erosion				+	+				+	+			+		+	+
Inflammation			+	+	+	+	+	+	+	+	+	+	+	+	+	+
Bile concretion				+									+			+
Mucous retention			+							+		+				+
Muscle hypertrophy			+	+	+			+		+	+	+	+		+	+

Although these changes have been extensively studied histologically, their clinical significance has not been generally recognized. A clinico-pathological investigation has, therefore, been undertaken of cases exhibiting fundal lesions which have been operated upon (by H. M. G.) during the period January, 1954—March, 1956, when 64 cholecystectomies or laparotomies for biliary tract disease were performed. Of these cholecystectomies, the presence of fundal changes was discovered at operation in 16 instances, giving an approximate incidence of 20 per cent. The clinical and pathological findings are summarized in *Table I*, whilst selected cases are included in the text.

CASE REPORTS

Cholecystitis Glandularis Proliferans Cystica (C.G.P.C.).—

Case 1.—A man of 53 years was admitted with jaundice of 8 weeks' duration. There was a history of flatulent dyspepsia for twelve years, accompanied by discomfort in the right hypochondrium. Liver and

spleen were not palpable, although the stools were clay-coloured. Shortly before operation, liver function tests showed serum bilirubin 8·4 mg./100 ml., serum alkaline phosphatase 22·2 King-Armstrong units, thymol turbidity less than 1, thymol flocculation 0, colloid gold test 0. At laparotomy there was found to be a fundal nodule in the wall of the gall-bladder. No gall-stones were palpated, but the bile-duct was greatly thickened. A diagnosis of

FIG. 421—*Case* 1. Cholangiogram through biliary drainage tube, showing small apical diverticulum.

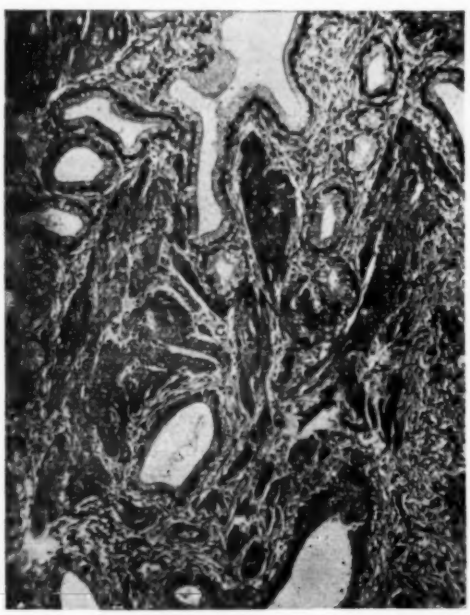

FIG. 422.—*Case* 3. Representative field from the apical lesion, showing proliferation of glandular and muscular elements. (× 110.)

stenosing cholangitis was made. The bile-duct was opened, the sphincter of Oddi dilated with a bougie, and a drain inserted into the bile-duct. A small piece of liver which was excised for histological examination showed the changes of chronic cholangitis, with some degree of retention of bile within the liver lobules. Cholangiogram showed the small apical diverticulum (*Fig.* 421). After the operation the jaundice gradually subsided.

Case 2.—A woman aged 63 years was subjected to bilateral transperitoneal adrenalectomy for advanced carcinoma of the breast. During the operation a fundal thickening of the gall-bladder was encountered as an incidental finding. No gall-stones were present. Although on subsequent questioning the patient denied having had gall-bladder symptoms, a cholecystogram which was performed after recovery from the operation showed a poorly functioning gall-bladder, which was reported as pathological by the radiologist.

Case 3.—A woman of 67 gave a history of intermittent dyspepsia and abdominal pain with episodes of colic for three years. There was tenderness and guarding below the right costal margin. At operation a large gall-bladder was found with a thick-walled fundal diverticulum 2 cm. in length, histologically showing cholecystitis glandularis proliferans (*Fig.* 422). Stones were found in the biliary tract at operation. Dilatation of the sphincter of Oddi, which was found to be stenosed, was performed.

Case 4.—A woman of 61 years complained of recurrent pain in the right hypochondrium and nausea for some years, together with flatulence, aggravated by fatty foods. These symptoms had become more frequent and more severe during the last six months. Bowel action had been regular and no melæna had been noted. Apart from a positive Murphy's sign, physical examination revealed nothing of significance. It was thought that her symptoms were due to gall-bladder disease. At operation a carcinoma of the transverse colon was found with multiple metastases in the liver. A right hemicolectomy was performed. A thickened diverticulum was found at the apex of the gall-bladder and cholecystectomy was carried out. The apex of the gall-bladder

FIG. 423.—*Case* 4. Formation of a biliary concretion, containing cholesterol clefts, in an isolated acinus of the apical lesion, which is surrounded by fibrous tissue. (× 110.)

showed histologically the characteristic changes of cholecystitis glandularis proliferans cystica, with minute biliary concretions within the lumen (*Fig.* 423), in addition to a widespread chronic inflammatory cellular reaction.

Case 5.—A woman aged 67 was investigated early in 1953 for symptoms thought to be related to gall-bladder

disease. A normal cholecystogram was obtained. Two years later, since symptoms persisted, this investigation was repeated and a poorly concentrating gall-bladder was demonstrated radiologically. At operation a fundal thickening was found, the gall-bladder containing numerous small pigment stones. Histological examination showed chronic inflammatory changes with apical cholecystitis glandularis proliferans cystica. Episodes of severe biliary colic occurred for a period of three months following operation.

Fundal Diverticula.—

Case 6.—A woman of 66 years of age gave a history of attacks of flatulent dyspepsia, vomiting, and right-sided para-umbilical pain. On examination there was tenderness in the right hypochondrium, a positive Murphy's sign, and slight tenderness in the right iliac fossa. Radiologically, no abnormality of stomach or duodenum was detected, the appendix later filling normally with barium. Cholecystography showed good filling of the gall-bladder and normal contraction after a fatty meal. The apical portion of the gall-bladder appeared to be separated from the rest by a constriction. At operation a thick-walled apical diverticulum was found in an otherwise apparently normal gall-bladder. There were no stones. Cholecystectomy was followed by relief of symptoms. Histologically there was evidence of chronic inflammation in the body of the gall-bladder, whilst the wall of the diverticulum showed the characteristic mucosal and muscular changes of cholecystitis glandularis proliferans cystica.

FIG. 424.—*Case* 8. Cholecystogram, showing apical diverticulum only.

Case 7.—A woman aged 47 had experienced flatulent dyspepsia, vomiting, and right-sided para-umbilical pain for two years. There was tenderness in the right hypochondrium and right iliac fossa, with a positive Murphy's sign. A barium meal showed no abnormality of the stomach, duodenum, and appendix, and a barium enema also gave a negative result. The cholecystogram was reported as follows: "The gall bladder is seen to be well filled and free of negative shadows. It is even in outline except for a shadow near its fundus which appears to be cut off from the main portion of the gall-bladder shadow. The gall-bladder contracts down normally after a fatty meal." No abnormality was found at laparotomy apart from the diverticulum of the gall-bladder. Cholecystectomy was performed, which cured the symptoms. The wall of the diverticulum was considerably thicker than the rest of the gall-bladder, which appeared otherwise normal. A narrow opening separated the diverticulum from the main cavity of the gall-bladder. There were no gall-stones. Histologically, appearances in the fundal portion were typical of cholecystitis glandularis proliferans, with a moderately severe degree of chronic inflammatory cellular infiltration throughout the gall-bladder.

FIG. 425.—*Case* 8. Cholecystogram, 18 mth. later. The presence of calculi is now evident.

Case 8.—A woman aged 28 years first felt epigastric pains, spreading to the back and shoulders, a fortnight after the birth of her first baby. These attacks were not related to eating fatty foods. She had noticed that during an attack her urine became frothy and dark brown in colour. Episodes of pain recurred every two or three weeks. Apart from slight epigastric tenderness, there were no abnormal physical signs. Barium meal showed no abnormality, whilst liver function tests and serum amylase levels were normal. On cholecystography the gall-bladder concentrated the dye well and contracted normally after a fatty meal. No calculi were seen (*Fig.* 424). There was a diverticulum of the fundus of the gall-bladder, which was seen to be cut off completely from the main shadow in oblique views. In spite of a gall-bladder diet and antispasmodics, symptoms continued. In addition to her previous attacks, she developed occasional attacks of severe pain across the upper abdomen, associated with vomiting, which left her weak and exhausted. A further cholecystogram, 18 months after the first radiograph, showed a well functioning gall-bladder with the same fundal pattern. This time a number of gall-stones were shown up in the gall-bladder (*Fig.* 425).

At laparotomy the pancreas was found to be enlarged and indurated. A thickened gall-bladder with a fundal diverticulum and containing several mixed stones was removed. The bile-duct was opened and the sphincter of Oddi was dilated. It yielded to dilatation like a fibrotic stricture. Histological examination showed active chronic inflammation, with fibrosis of cystic duct, gall-bladder, and diverticulum. The wall, both of the body of the viscus and of the diverticulum, showed some degree of muscular hypertrophy, with mucosal ectopia in the latter situation.

DISCUSSION

Cholecystitis Glandularis Proliferans Cystica.—In our series of 64 cases with gall-bladder disease there were 16 instances of fundal lesions, the latter most frequently taking the form of a nodule showing the histological changes of cholecystitis glandularis proliferans. Differences of opinion exist regarding the mode of production of this pathological change, which is characterized by a disorganization of the muscularis and by the presence of pockets of actively secreting mucous membrane throughout the thickened apical region of the gall-bladder (*Fig.* 426). Our results suggest that the lesion does not occur unless the function of the gall-bladder is already disturbed; an inflammatory cellular reaction throughout the wall of the gall-bladder has been an almost invariable finding in our cases. These inflammatory changes may play some part in the production of the lesion by causing œdema of the wall of the viscus and weakening of the muscle bundles, thus allowing the extrusion of epithelium beyond the thin muscularis. The force which drives the epithelium into and through the muscularis must be derived from increased intracystic pressure, due to biliary obstruction or dyskinesia. Apart from cholecystitis or calculi in the gall-bladder, cystic duct, or bile-duct, stenosing cholangitis, stricture and spasm of the sphincter of Oddi, and pancreatitis were found as associated lesions in our cases.

The demonstration of the presence of a small fundal diverticulum radiologically, or of a fundal nodule at laparotomy may therefore be taken as a diagnostic sign of disordered biliary function. Since gall-stones may fail to show up on cholecystography or may be too small to be palpated in a distended gall-bladder, this sign may be of comparable diagnostic value, or of even greater significance than the finding of a gall-stone. The fundal nodule may be present before any other gall-bladder pathology is obvious.

In those cases where the increased intracystic pressure, which is one of the factors in the production of the fundal nodule, is the consequence of disease of the gall-bladder or cystic duct, cholecystectomy alone will cure the symptoms. If, on the other hand, there is neither obvious inflammation of the gall-bladder, nor obstruction to the flow of the bile into the bile-duct, the evidence of biliary dyskinesia, supplied by the presence of the fundal diverticulum, should draw the attention of the surgeon to the sphincter of Oddi. Unless the obstruction at that site is overcome, post-cholecystectomy symptoms may persist. Pain may be more severe than it was before the operation, since the rising pressure inside the bile-ducts can no longer be lowered by the expansion and dehydrating mechanism of the gall-bladder (McMaster and Elman, 1926; and see our *Case* 5).

Gall-stones were found in the biliary passages in 9 of our 16 cases, whilst biliary concretions were seen within the ectopic mucosal acini on several occasions. As in our *Case* 4, these little stones may occur in the absence of calculi elsewhere in the biliary tract. These findings raise the question of whether the presence of cholecystitis glandularis proliferans may not predispose to the formation of biliary calculi in some instances.

FIG. 426.—Diagram to show the nature of cholecystitis glandularis proliferans cystica.

Fundal Diverticula.—Although a diverticulum may occur in any part of the gall-bladder, it is most commonly seen at the fundus. The formation of such a lesion may be explained either on embryological grounds by incomplete absorption of the once solid interior of the gall-bladder (Weltz, 1894; Rukstinat, 1936; Lockwood, 1948) or, as a consequence of raised intravesical pressure, by enlargement of a Rokitansky–Aschoff sinus. A congenitally derived diverticulum might be expected to have a normally contractile wall and should not therefore predispose to the occurrence of pathological changes. A diverticulum which is produced by the enormous distension of an R–A sinus will probably be found in association with other lesions. The latter type of diverticulum is characteristically narrow necked and contains fewer muscle-fibres than the body of the gall-bladder. Calculi which have once found their way into such a diverticulum will not be expelled again into the lumen of the gall-bladder.

The phrygian cap deformity, commonly seen on cholecystography, is thought to denote the existence

of a congenital diverticulum, which is as a rule of no pathological significance (Boyden, 1935; Meyer, Carter, and Mecker, 1937). Lockwood, however, found a significantly raised incidence of gall-bladder disease in these patients. Occasionally appearances are found where a band seems to separate the fundal portion completely, or almost completely, from the rest of the gall-bladder. Oblique views may be necessary for its clear demonstration. Such cases are likely to be examples of diverticula formed from R–A sinuses, which have penetrated the muscularis before expanding to form a pouch under the serous coat.

Cases are not infrequent in which the association of fundal diverticula with other obvious pathology, such as calculi, will draw the attention of the diagnostician to the gall-bladder. Inflammatory changes in the viscus in these cases often interfere with its concentrating power of dye and overshadow the diverticulum in diagnostic significance. In Rukstinat's 2 cases the viscus gave a poor shadow only. Cases with fistulæ from a fundal diverticulum leading to various sites have been reported—skin (Toida, 1913), stomach (Shenig and Schondube, 1925), jejunum (Pertl, 1933)—which must have given obvious signs of the underlying inflammation.

Cases such as that reported by Beaumont (1950) in which, following biliary colic, a fundal diverticulum was found in an otherwise normal gall-bladder, must be unusual. In our examples of this condition there has been histological evidence of chronic inflammation of the body of the gall-bladder, whilst the mucosal changes of cholecystitis glandularis proliferans cystica in the walls of these diverticula make their derivation from R–A sinuses appear likely. In *Case* 8, calculi developed during the period of observation. The progression towards inflammation and calculus formation is similar to that of cases with fundal nodules, such as *Case* 5.

The narrow-necked fundal diverticulum appears to have the same mode of formation as apical cholecystitis glandularis proliferans cystica and can be regarded as having the same clinical significance. Increased intravesical pressure is probably of greater importance than inflammation in the production of the larger diverticula, since inflammatory thickening of the wall would tend to prevent the excessive expansion of an R–A sinus. In those cases where the production of fistulæ has been reported, the increase in pressure appears to have been due to the presence of calculus in the cystic duct. Spasm or stenosis of the sphincter of Oddi may well be important aetiological factors in those cases where obstruction by calculus is not encountered.

SUMMARY

Of 64 consecutive cholecystectomies, 16 showed acquired apical gall-bladder lesions, recognizable at laparotomy as a fundal nodule or diverticulum and showing the histological changes of cholecystitis glandularis proliferans cystica. It is thought that the development of radiologically recognizable narrow-necked fundal diverticula may be part of the same pathological process, predisposing factors being intramural inflammatory changes and rises in intracystic pressure.

The presence of gall-stones, of a radiologically visible diverticulum, or of a palpable gall-bladder diverticulum, or fundal nodule at laparotomy, are findings each of approximately equal value in the diagnosis of biliary dysfunction. Where a fundal lesion is encountered, possible causes of rise in intracystic pressure should be sought and corrected, in order to avoid the occurrence of post-cholecystectomy symptoms.

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INTRATHORACIC DEVIATION OF A URETERIC LOOP

By A. H. SWITHINBANK, Liverpool

This communication describes and discusses a well-defined pathological entity which would appear to be of great rarity.

A case is reported in which the upper part of the right ureter was found to take an abnormal course, forming an intrathoracic loop with limbs passing through a posterolateral diaphragmatic aperture. The clinical features and distinctive radiological appearances are presented, together with an account of successful surgical treatment and a discussion of pathogenesis.

Fig. 427.—Pre-operative intravenous pyelograph showing intrathoracic deviation of a loop of the right ureter. Right paravertebral soft-tissue shadow is seen.

Fig. 428.—Retrograde pyelograph in a right posterior oblique view, showing extent of intrathoracic deviation of the right ureteric loop.

CASE REPORT

Mrs. M. K., a housewife, aged 60 years, was admitted to Liverpool Stanley Hospital in July, 1956, with a history of attacks of right-sided pain since June, 1955.

The pain, in each attack, began abruptly below the right scapula at a point well localized by the patient, about 3 in. from the midline, and just above the level of the tenth rib. Between half to one hour after onset it radiated downwards and forwards, mainly to the right groin, but also to the right hypochondrium. There was no shoulder-tip pain. The duration of an attack varied from four to twenty-four hours and the pain was always most marked in the right subscapular region. The intensity of pain varied somewhat during an attack, but usually reached a severe degree at some stage and required morphia for its partial relief. It was not described as colicky. The end of an attack was as abrupt as the onset. For a few weeks before admission, bouts of pain occurred approximately once a week. Before this period there were intervals of freedom up to five weeks in duration. A desire to micturate accompanied each episode, but frequent passage of small amounts of urine did not give any relief. The more severe attacks were associated with vomiting of any recent food, followed by bilious retching. It was noticed that a deep sigh increased the pain, but coughing and straining were without effect in this respect. After admission the patient was seen in an attack conforming to the description given.

In April, 1955, cholecystectomy through a Kocher's incision had been carried out at the Liverpool Stanley Hospital. This was undertaken for a ten-months' history of intermittent pain and flatulence which appeared to be characteristic of chronic cholecystitis. The pain started and was maximal in the right hypochondrium, and radiated to the back on the right side. A Graham's X-ray examination showed the presence of a gall-stone. At operation the gall-bladder, containing a solitary cholesterol stone, was found to be thickened, with some omental adhesions. The patient was certain that the pain for which cholecystectomy was undertaken was different from her subsequent complaint. Cholecystectomy was followed by a post-operative bronchitis.

Before this illness there had been no right-sided pain, and the only feature with possible relevancy was an accident at the age of 7 years in which the patient was run over by a horse-drawn lorry. She thought that the vehicle had passed over the middle of her body and that, as a result, she had been in hospital for some days. There had been no apparent residual disability from this accident.

On Examination.—This showed no cause for the symptoms. The patient appeared healthy and there was

no evidence of pulmonary or cardiac disease. There was a sound right subcostal scar in a fat abdominal wall. Blood-pressure 170/100. Blood and urine examinations showed no abnormality. Blood-urea was 28 mg.

FIG. 429.—Post-operative intravenous pyelograph showing the corrected course of the right ureter.

per cent. Blood-group O, Rh positive. Cystoscopic appearances were normal.

The decisive investigation was radiological. X-ray examination of the diaphragmatic region showed a right paravertebral soft-tissue shadow from the level of the eighth to eleventh thoracic vertebræ.

Intravenous pyelography confirmed good renal function on both sides, and apparent abnormality was restricted to the upper right urinary tract (*Fig.* 427). The right ureter pursued a most unusual course, forming a loop passing above the diaphragmatic level. The limbs of the loop lay together, an appearance suggesting that they passed through a narrow strait between abdomen and thorax. The right kidney occupied its normal position, the dilated ureter running upwards and medially from the renal pelvis.

Right retrograde pyelography (*Fig.* 428) confirmed the unusual course of the ureter and showed an early degree of hydronephrosis with some dilatation of the proximal ureter.

General radiography of the chest and gastro-intestinal tract gave no further help.

Osteo-arthritic changes were present in the spine, most marked in the lumbar region.

In the light of these findings reference was made to the series of radiographs taken by Graham's method early in 1955 : the paravertebral soft-tissue shadow could be distinguished, not very distinctly, in one of the films.

The radiological findings were considered due to an abnormality in which the right ureter passed through a pleuroperitoneal hiatus of the diaphragm (foramen of Bochdalek) on the right side. The nature of the paravertebral soft-tissue shadow remained a matter for speculation. Operation was indicated by the evident relationship between the symptoms and the ureteric abnormality.

AT OPERATION.—This was carried out by Mr. S. V. Unsworth (July 7, 1956) with general anæsthesia. Incision was oblique in the right loin, extending up on the back of the chest wall. The greater part of the twelfth rib and part of the eleventh rib were removed. The right kidney was found in the usual position. The lower pole contained two small, simple cysts, and there was a minor degree of external hydronephrosis. From the renal pelvis a dilated ureter took a somewhat tortuous course upwards and medially, passed through an aperture in the diaphragm, and returned to the abdomen through the same opening. This opening was a well defined slit-like gap between bundles of diaphragmatic muscle. It lay 4 cm. above the middle of the right medial arcuate ligament, was 2·5 cm. in length, and contained some extra-peritoneal fat in addition to ureter. It was possible to introduce a finger and bring the ureteric loop into the abdomen, the loop being but loosely attached, without substantial vascular connexion in the chest. Digital examination revealed nothing but fat in the walls of the cavity from which the ureteric loop had been delivered.

The hiatus was closed with four braided terylene sutures. The simple cysts of kidney were 'uncapped' and their remaining walls were carbolized. The peritoneal and pleural cavities were not opened during the operation. The wound was closed with a corrugated rubber drain down to the kidney.

POST-OPERATIVE PROGRESS.—This was uneventful, and the patient has remained free from symptoms. Intravenous pyelography, three weeks after operation, showed the right ureter pursuing a more normal, sub-diaphragmatic course, with some redundancy, but no marked dilatation (*Fig.* 429). The appearance of the right renal pelvis was normal. The paravertebral soft-tissue shadow showed no appreciable change.

DISCUSSION

Intermittent obstruction of the right ureter was the physiological abnormality responsible for the clinical features. Delay in recognizing this from the patient's history is explained, but not perhaps excused, by the occurrence of symptoms, soon after cholecystectomy for right-sided abdominal pain, in a patient whose spinal osteo-arthritis was a possible source of referred pain. In retrospect, two features of the painful attacks are of interesting significance: the high origin ; and exacerbation by a sigh, which involves a deep inspiration. It must be admitted that the precipitating cause of each painful attack remains obscure.

The level of ureteric obstruction, and the unusual anatomical variation responsible, were clearly indicated by the radiographs. Incidentally, it became evident that the paravertebral soft-tissue shadow in radiographs was due to fat outlined by relatively translucent lung tissue.

It remains to consider the pathogenesis of an abnormality with two principal components : a right posterolateral diaphragmatic hiatus ; and deviation, through this hiatus, of a loop of ureter.

No previous account of the abnormality has been discovered. There are a number of reported cases of intrathoracic ectopy of the kidney associated with a posterolateral diaphragmatic defect—an unusual condition. Spillane and Prather (1949), for example, report a case on the left side ; Fleischner, Robins, and Abrams (1950) report a case on the right side.

The cases described by these authors are of undoubted congenital origin and, with other reports, offer important evidence that the upper end of ureter on either side may, albeit in company with

the kidney, occupy a supradiaphragmatic position as a result of abnormal development involving an associated diaphragmatic defect. Spillane and Prather remark: "In order to explain the supradiaphragmatic kidney on an embryological basis, we are forced to hypothecate not only a high vascular origin but also retroperitoneal herniation through the pleuroperitoneal hiatus, which later closed sufficiently to incarcerate the kidney and prevented further herniation of other organs."

In the present case it is necessary to examine the possibilities of congenital and acquired factors in causation.

A history of a crush injury at the age of 7 years raises the question of trauma as a cause for the diaphragmatic aperture. It is difficult, however, to see how a length of normal ureter could rise into the chest, either at the time of the accident, which would involve a sudden elongation, or later. The ureter has a definite attachment to the posterior parietal peritoneum by connective tissue which serves as a tunica adventitia for the extensive peri-ureteric venous plexus, and it would require a singularly well localized and persistent force to cause its extension and elongation.

The same consideration obtains as an objection to the hypothesis that ureteric displacement might take place as an acquired extrusion through a congenital diaphragmatic aperture; and it seems reasonable to regard the ureteric deflection as due, primarily, to abnormal intra-uterine development.

In considering the embryology of the present case, emphasis may be placed on the relatively high position of the ureteric loop. If this is explicable, it is understandable that diaphragmatic 'descent' could be associated with incomplete closure, around the loop, of developing diaphragmatic elements which normally fuse in the right pleuroperitoneal region by the second month (20 mm. embryo stage). Although it is convenient here to visualize the diaphragm as descending to close around a segment of ureter, it should be mentioned that the final relationships of diaphragm, kidney, and upper ureter are determined, not so much by active growth and movement, as by relative shift of their rudiments incidental to changes in the developing body wall. Case reports of intrathoracic kidney demonstrate that this normal process may be modified so that the upper end of ureter (and kidney) finally lie above the definitive diaphragmatic level, and some reports describe abnormally high vascular attachment of the ectopic kidney.

Without help from the operative findings, reasons for high fixation of a segment of the right ureter, during the critical 17–20 mm. stages of the embryo, must be hypothetical; but in the absence of any other communication it may be worth while to speculate briefly on this point.

The venous drainage of upper ureter and kidney on the right side is intimately associated in early development with the rich and complex system in which the lower part of the vena azygos and adjoining inferior vena cava are consolidated. An unusually close and persistent association between the upper part of this system and the venous drainage of a segment of upper right ureter would seem to provide the likeliest explanation for relatively high fixation of a segment of the ureter during the 17–20 mm. stages of the embryo.

It may be remarked that an abnormal phase of venous, rather than arterial, development is the probable mediator in pathogenesis, because of the complexity and plasticity of the primitive venous bed in the region under consideration (as exemplified by frequent variation in the origin of vena azygos).

This hypothesis is not incompatible with the normal development of derivatives from the anlagen of kidney and ureter apart from the abnormally involved segment of ureter; nor is it seriously compromised by the absence, at operation, of significant intrathoracic ureteric vascular connexions—the ureteric vascular attachments are distinct from those of the kidney and normally assume their definitive adult pattern relatively late, with no persisting evidence of antecedent stages.

Although an embryological fault is suggested as the essential factor in pathogenesis, it is understandable, and even to be expected, that a gradual upward extension of the ureteric loop would take place as a result of differences between intrathoracic and intra-abdominal pressures. Such a gradually extending herniation of the loop (loosely attached in this instance), together with extraperitoneal fat, would help to explain delay in the onset of symptoms until later life. The operation of cholecystectomy was followed by bronchitis, and it is tempting to regard the associated episodes of raised intra-abdominal pressure as precipitating symptoms of ureteric obstruction by causing further extrusion of extraperitoneal fat through the narrow diaphragmatic hiatus.

SUMMARY

The occurrence is reported of intrathoracic deviation of a loop of the upper part of the right ureter through a posterolateral aperture in the diaphragm.

Clinical features and an account of successful surgical treatment are described.

It is suggested that the abnormality was primarily the result of abnormal intra-uterine development.

This is believed to be the first recorded instance of the condition.

My thanks for help in the preparation of this paper are due to Mr. S. V. Unsworth; Dr. Tudor J. Jones and Dr. E. W. Macmillan of the Department of Anatomy, University of Liverpool, who kindly advised on anatomical and embryological aspects; and Dr. G. Scarrow of the Department of Radiology, Liverpool Stanley Hospital.

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EXCISION OF THE GERMINAL MATRIX: A UNIFIED TREATMENT FOR EMBEDDED TOE-NAIL AND ONYCHOGRYPHOSIS

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A LOGICAL, simple, and effective operation for the embedded toe-nail was described by Quénu in 1887. This operation has not received the attention it merits. My experience of treating 100 toe-nails by excision of the germinal matrix has proved that, with a few exceptions, it provides an ideal treatment for both the embedded nail and onychogryphosis.

SURGICAL ANATOMY

The nail* is a horny plate of modified skin epithelium. The sides are parallel and the proximal edge is concave. The root of the nail is covered by the nail-fold and the sides of the nail are overlapped by nail-walls (*Fig.* 430). The horny thickening of skin at the edge of the nail-fold is called the eponychium or cuticle. At its distal end the nail overlaps the skin, and the horny thickening beneath the free border of the nail is called the hyponychium. Beneath the nail is a softer epithelial layer called the matrix or nail-bed which is continuous with the deeper or malpighian layer of the epidermis (*Fig.* 431). The matrix is divided into two parts.† That forming the lunula is called the germinal matrix because the nail grows from it. The remainder is called the sterile matrix and forms a surface over which the nail moves. The matrix is firmly attached to the underlying vascular connective tissue of the corium. When the nail is avulsed the thin sterile matrix is damaged and the vessels of the corium are exposed and bleed a little, but if the nail is separated from the matrix by sharp dissection no bleeding occurs.

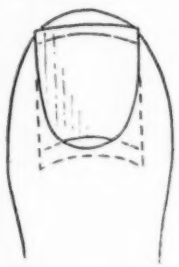

FIG. 430.—Diagram showing the relation of the nail to the nail-fold and the nail-walls.

It was Quénu who first proved experimentally that only the lunular matrix is nail-forming. He showed that if the nail is withdrawn and the lunular matrix excised the remaining matrix becomes cornified and covered with horny lamellæ, but does not form true nail-tissue. The lamellæ are easily separable instead of being welded into a solid plate and the cells, unlike those of the nail, contain no trace of nuclei. This cornification of the sterile matrix which occurs after excision of the germinal matrix may account for the persistence of the idea that the whole of the matrix is nail-forming. Further evidence that this layer of cornified epithelium is not true nail is provided by the fact that it does not have to be trimmed. The cells of the germinal matrix grow longitudinally whereas those of the sterile matrix grow only in thickness. Growth of the sterile matrix is normally no more than is necessary to maintain a surface on which the nail slides. It may contribute some cornified elements to the nail, but this addition is small because the nail does not appreciably thicken as it moves distally from the edge of the lunula (*Fig.* 431).

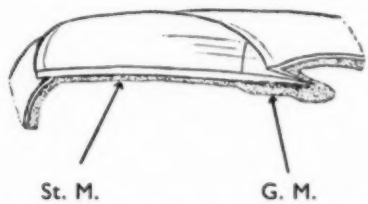

FIG. 431.—Diagram of a longitudinal section through the nail. The lunula is seen to correspond to the germinal matrix (G. M.). The rest of the nail-bed consists of the sterile matrix (St. M.). (*After W. E. Le Gros Clark in 'Tissues of the Body'.*)

SURGICAL PATHOLOGY

Embedded Toe-Nail.—The essential pathology is an ulceration of the nail-groove in the area of contact between the edge of the nail and the nail-wall. The swollen nail-wall overlaps the nail and gives the appearance of an ingrowing nail (*Fig.* 432). The term 'embedded' is preferred to the term 'ingrowing' because in nearly all cases the shape of the nail is normal. In any event, there can be no active ingrowing of the sides of the nail; the parallel longitudinal nail-ridges (*see Fig.* 430) are evidence that the nail grows straight out from the root.

There are three possible factors in the pathogenesis: (1) Pressure on the nail-walls; (2) Deformity of the nail; (3) Sweat and dirt. There is no general agreement on the relative importance of these factors. The following observations are based on a detailed study of 50 cases.

1. *Pressure on the Nail-walls.*—Patients with embedded toe-nails usually have fleshy toes with bulky nail-walls, but a history of abnormal pressure before embedding is uncommon. Twelve patients

* In some anatomical descriptions (*Gray's Anatomy*) the term 'nail' includes both the horny plate and the underlying epithelium or matrix. This terminology may suit the anatomist, but it will confuse the surgeon who, when he avulses the nail, expects to leave the matrix behind.

† In view of the fact that growth of the nail is now known to occur only from the lunular matrix, some writers restrict the term 'matrix' to this area and call the sterile matrix the nail-bed. This terminology is not universal and I prefer the unequivocal terms used by Le Gros Clark (1952).

in this series of 50 cases mentioned pressure of footwear, but only 4 were certain that this was the cause of the embedding. Two cases were due to tight plaster casts and 2 were due to pressure of bedclothes during prolonged recumbency.

A significant feature which has emerged from this study is the fact that in 42 out of 50 toe-nails the outer side of the nail-plate was either the first or the only side to become embedded. This relatively high incidence of embedding of the outer side of the nail

FIG. 432.—Showing the part played by the second toe in embedding of the outer side of the nail.

must be due to the effects of pressure on the toe. On the inner side pressure of footwear is spread over the whole inner border of the hallux, whereas on the outer side this pressure is localized to the area of contact with the second toe (*Fig.* 432).

The fact that embedding of the toe-nail is rare in women who wear narrow, pointed shoes is evidence that pressure of footwear is not the only factor. On the other hand, embedding is almost unknown in barefooted races and evidence will be adduced to show that footwear chiefly determines the other factors in the pathogenesis.

2. *Deformity of the Nail.*—There is a wide variation in the normal shape of the nail and the nail-walls are adaptable to very marked curvatures in the nail. In fact, thick nails with rolled-in edges are less likely to give rise to trouble than flat nails with sharp edges abutting against the nail-walls (*Fig.* 432). Three types of nail deformity were encountered in this series :—

1. Two patients had excessive incurving and embedding due to shoe pressure. The nail deformity was due to prolonged pressure on the nail-plate.

2. Two patients had onychogryphotic nails which had become embedded. With advancing years the curvature of the nails tends to increase and hypertrophy of the nails is common, but embedding is rare because at this age there is a corresponding atrophy of the nail-walls.

3. Self-inflicted nail deformity was common and is probably an important factor in the pathogenesis. The prevalent habit of cutting the nails short leaves the pulp unsupported at the end of the toe and weight-bearing presses the soft tissues into and around the nail-edges (*Fig.* 433). This effect is enhanced by the fact that cutting the nails short frequently results in the retention of a spike of nail in the nail-groove. Thirty-seven patients said they had always cut their toe-nails short and 4 of these had the habit of tearing the strips of nail off with their fingers. Twenty-one admitted that vigorous cutting had either caused or aggravated their condition.

In 7 patients embedding followed injury. Five of these injuries resulted in loss of the toe-nail and the new nail became embedded in spite of the fact that it was not deformed. This illustrates a similar

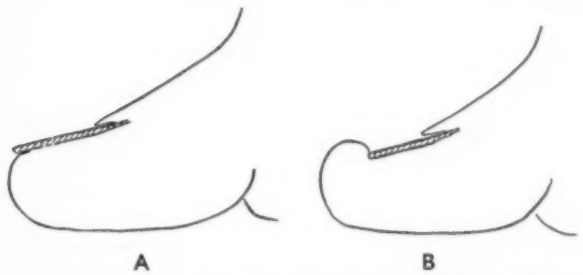

FIG. 433.—A, The nail normally supports the pulp during weight-bearing. B, If the nail is cut short the unsupported pulp is pressed around the sides of the nail.

mechanism ; the advancing edge of the new nail becomes buried in the same way as a nail which has been cut too short (*Fig.* 433). This also explains why avulsion of the nail is nearly always followed by recurrence and often causes aggravation of the embedding.

3. *Sweat and Dirt.*—Retained sweat leads to maceration of the nail-groove and this, together with dirt, will predispose to ulceration and infection. Twenty-six patients said that they suffered from excessive sweating of the feet. Clark and Dillinger (1947) reported an annual incidence of embedding of 11·3 per cent in infantrymen in the tropics, as against 2·9 per cent in sailors. This illustrates the

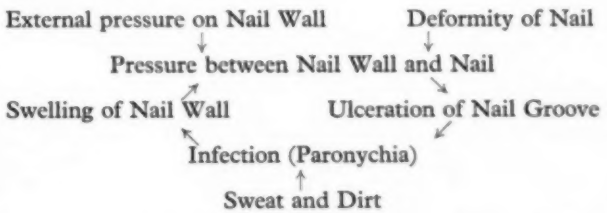

FIG. 434.—Pathogenesis of embedding ; a vicious circle mechanism.

combined effect of heavy footwear, hard use of the feet, and excessive sweating.

It will now be evident that footwear, in the form of boots, shoes, and hosiery, is responsible for the three most important factors in the pathogenesis of the embedded nail, namely, crowding together of toes, cutting of toe-nails, and sweat retention.

In all severe cases infection dominates the picture and makes the condition self-perpetuating. Once infection is established a vicious circle comes into operation and the condition will persist in the absence

of the original causes (*Fig.* 434). Infection leads to swelling of the nail-wall; this increases the pressure on the nail, which in turn causes further ulceration and infection.

The fully established case presents as a severe paronychia and it may be difficult or impossible to determine the original causes of the embedding. This is one reason for the differences of opinion among writers on the subject; some emphasize the importance of pressure on the nail-wall and others

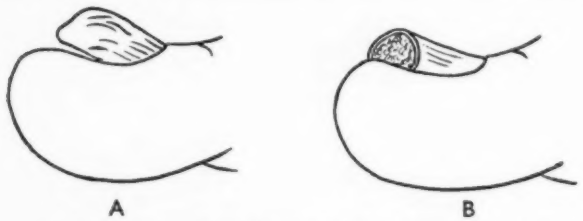

FIG. 434.—Onychogryphosis may be due to A, Thickening of the nail; or B, Thickening of the sterile matrix. Combined types occur frequently.

the abnormalities in the nail. All, however, are agreed that it is the addition of infection which induces the patient to seek treatment.

Onychogryphosis.—There are two main types of onychogryphosis :—

1. Thickening of the nail due to disordered growth of the germinal matrix (*Fig.* 435 A). This may occur at all ages; in the young it usually follows trauma.

2. Thickening of the sterile matrix causing elevation of the nail by horny lamellæ (*Fig.* 435 B). This is a common senile change and may be regarded as a transgression of the hyponychium beneath the body of the nail.

These two types merge into one another; in many cases there is a combined thickening of both the nail and the sterile matrix. This is characteristic of onychogryphosis due to fungus infections.

The distinction between the sterile and germinal matrix is always preserved in the nail-bed of onychogryphosis. The lunula is not seen in the nail, but after avulsion of the nail the lunular matrix is clearly distinguishable by its smoother and paler surface (*Fig.* 436 B). The germinal matrix is frequently hypertrophied and occupies a greater proportion of the nail-bed than in the normal nail, but the functions of the two parts of the matrix continue true to type. Nail-tissue is only formed from the germinal matrix. The sterile matrix may heap up cornified epithelium, but it never forms true nail-tissue.

TREATMENT

Conservative Treatment.—Two types of conservative treatment are available for the embedded nail: (1) The nail is allowed to grow long and elevated above the soft parts. This is the rational approach and may lead to a cure. (2) The embedded nail is cut away from the nail-groove. Temporary relief may be gained by this manœuvre, but this treatment perpetuates one of the causes of embedding. Many patients who seek surgical treatment have continued to cut away the 'ingrowing' nail until it becomes too deeply embedded and too painful to continue.

Simple avulsion of the nail is often done in the hope that it will lead to a cure, but relapse is frequent. In onychogryphosis the deformity invariably recurs after avulsion. Following removal of an embedded nail, infection subsides and the ulcer heals, but the scar is usually too weak to withstand the advancing edge of the new nail.

Removal of the buried nail is, however, an essential preliminary to any operation for a septic embedded nail. The whole length of the embedded side of the nail should be removed in order to allow time for the infection to subside before the new nail grows again into the groove. I now wait 4 weeks before proceeding to the removal of the germinal matrix. The only post-operative sepsis in this series occurred when I operated too soon after removing the nail from an infected toe.

Operation for Bilateral Embedded Nail and Onychogryphosis.—Ring block anæsthesia is employed using 2 per cent lignocaine without adrenaline. A tourniquet is essential because the germinal matrix must be seen and carefully dissected out. Two $\frac{1}{2}$-in. incisions are made which begin at the junction of the nail-fold and nail-walls (*Fig.* 436 A). Three flaps are dissected which consist of skin only; the layer of epithelium on the deep surfaces of the nail-fold and nail-walls is left in situ (*Fig.* 436 B, C). At this stage the nail is removed, unless this has already been done to eradicate sepsis.

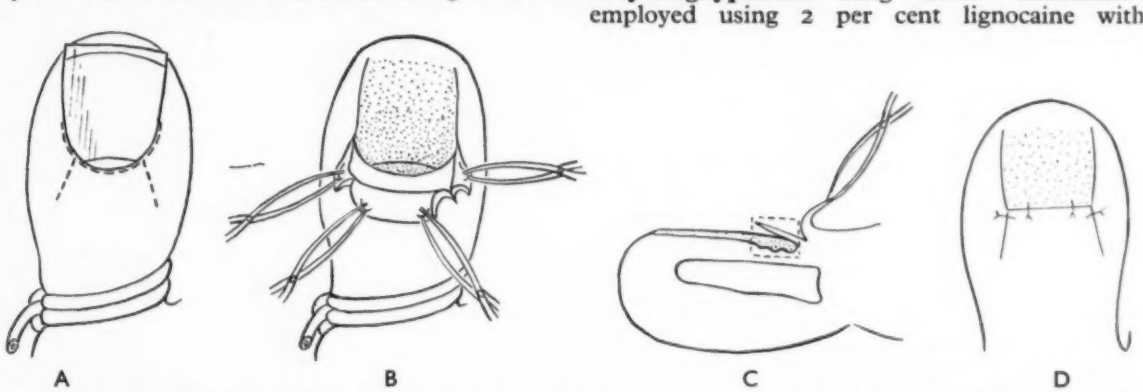

FIG. 435.—Technique of total excision of the germinal matrix. A, Skin incisions. B, Skin-flaps dissected and retracted. The epithelial layer on the deep surface of the flaps is left attached to the matrix. C, Longitudinal section showing the block of tissue to be removed. D, Skin-flaps sutured to cover the raw area.

The skin-flaps are retracted and a block of tissue is dissected out which consists of the germinal matrix and the overlying deep layer of the nail-fold and walls (*Fig.* 436 C). The skin-flaps are stitched back to cover the raw area (*Fig.* 436 D). The central flap can usually be approximated to the cut edge of the

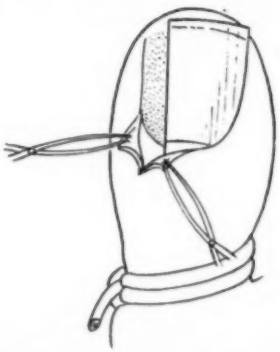

FIG. 437.—Segmental excision of the germinal matrix. The skin-flaps have been raised preparatory to excision of a segment of the germinal matrix.

removed it is examined; it should consist of an intact epithelial pocket. It is unnecessary to cut away or scrape the periosteum; the matrix is separated from the bone by a thick layer of corium (*see Fig.* 436 C).

In my early cases I cut along the line of the convex edge of the lunula and this resulted in slight regeneration of nail in some patients. Evidently microscopic areas of germinal matrix are likely to be left behind unless the knife is kept clear of the macroscopic line of demarcation. It is easy to identify the germinal matrix immediately after the nail has been removed; it is smoother and paler than the sterile matrix. This distinction may be obscured if the nail has recently been avulsed, in which event an estimate of the extent of the lunular area will have to be made.

The epithelium on the deep surface of the nail-fold and nail-walls is removed because unless this is done an epithelial pocket will be formed when the flaps are sewn back over the raw area. This delays healing.

After removal of the tourniquet there is brisk hæmorrhage which is controlled by a pressure

A B

FIG. 438.—Post-operative appearances. A, After total excision. B, After segmental excision.

sterile matrix without difficulty; if it cannot, a small gap can be left which heals rapidly.

Operation for Unilateral Embedded Nail.—The technique is similar in all essentials to the above operation except that the excision of the germinal matrix is limited to a segment ¼ in. wide at the corner of the nail. One skin incision is used and two flaps are raised in order to expose the germinal matrix of the affected side of the nail (*Fig.* 437). The raw area is completely covered by the skin-flaps.

Certain details of technique need emphasis. The operations must be exact dissections to the limits of the germinal matrix. Distally the matrix is divided just beyond the edge of the lunular area, proximally the dissection reaches nearly to the joint line, and laterally it continues until the knife reaches the fatty tissue of the pulp. In the operation for total excision of the germinal matrix the tissue is excised in two parts by starting from the midline and working to each side. As each block of tissue is

dressing. On the third day a small dressing is applied and the patient is allowed to walk in soft shoes or slippers. All the patients in this series were treated as out-patients except for a few who were admitted to hospital for social reasons.

RESULTS

I am reporting the results of 50 operations on embedded nails and 50 operations for onychogryphosis. All the patients have been followed up for 6 months or longer, 66 were followed up for a year or longer, and 15 for 3–5 years. Typical appearances after operation are shown in *Fig.* 438.

Embedded Toe-nail.—There were 32 total excisions of the germinal matrix, 17 segmental excisions, and one patient in whom segmental excision was done on both sides of the nail. All these operations have been successful. In 5 cases there was regeneration of nail-tissue, but only sufficient to form thin flaky nails which caused no symptoms.

Onychogryphosis.—There were 3 recurrences of nail-growth, but only one gave rise to symptoms and required a second operation. Several patients with residual thickening of the sterile matrix thought that the operation was only partly successful until it was pointed out that the scales could be removed

FIG. 439.—Excessive upward curve of the pulp after excision of the nail in a patient with hallux rigidus.

from the nail-bed by gentle scraping. It is therefore advisable to warn patients beforehand of this possibility.

By adding the two groups together I am able to report 100 operations with one failure. Regeneration of nail-tissue occurred in the early cases before I had realized that the knife must be kept clear of the edge of the lunula when excising the germinal matrix. In the last 70 cases there has been no recurrence of

FIG. 441.—The terminal amputation operation showing recurrence of nail-growth on the left side.

nail-growth. A 6-months' follow-up is considered adequate because 7 out of the 8 nail-tissue regenerations occurred within that time. In one patient the time taken to regenerate was not determined, but it was definitely less than a year.

The follow-up revealed 4 patients who might have been more comfortable if they had had amputation of the end of the toe. Two of these patients had hallux rigidus and a stiff interphalangeal joint. Shortening of the toe would have relieved strain on the metatarsophalangeal joint (Pridie, 1955). One had hyperextension deformity of the interphalangeal joint due to hallux rigidus, and another a valgus deformity of the interphalangeal joint. Removal of the nail exaggerated the deformity of the terminal phalanx by allowing the pulp of the toe to become distorted (*Fig.* 439). Both these patients had mild discomfort from pressure over the tip of the toe.

DISCUSSION

Quénu originated total excision of the germinal matrix in 1887. This operation was rediscovered by Wilson (1944), by Zadik (1950), and by myself. It is not surprising that such a fundamentally sound operation should be rediscovered several times and

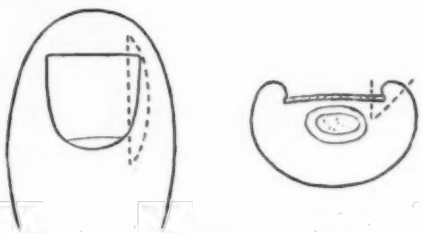

FIG. 440.—Drawings copied from a description of the classical wedge excision operation. Exposure of the germinal matrix is inadequate.

it is probable that it will continue to be rediscovered until it becomes a standard procedure in our text-books of surgery.

Segmental excision of the germinal matrix was also described by Quénu, but he did not advocate it because the fragment was too small. This difficulty would not have arisen if a tourniquet had been used. In my experience the segmental operation is satisfactory in selected cases; many embedded nails have swelling of both nail-walls and when in doubt total excision should be done. Segmental excision was described by Heifetz (1945), who emphasized the importance of using a sharp scalpel and making a careful dissection of the germinal matrix.

In an earlier paper Heifetz (1937) advised curettage of the germinal matrix. Whitesell (1945) and Manson (1946) also used the curette for this purpose. Others have advocated curetting or coring out a strip which includes both the germinal and sterile matrix (Winograd, 1929; Steinberg, 1954). The matrix is a firm, tough epithelial structure which is totally unsuited to the use of the curette. No surgeon would advocate curettage in order to eradicate a dermoid or sebaceous cyst; it would be an invitation to recurrence. If the bad reputation of toe-nail surgery is to be blotted out, then the curette must be laid aside and a sharp knife used to make an exact dissection of a well-defined structure.

Operations for embedded toe-nail may be designed to remove the nail from the nail-wall or to remove the nail-wall from the nail. Many surgeons have pointed out that because the chief cause of embedding is pressure of the soft tissues against the nail, therefore the logical treatment is to eliminate the nail-wall. Cotting (1873) sliced away the nail-wall and side of the toe and left the wound to granulate. Others (Howard, 1893; Ney, 1923) took wedges or slices out of the side of the toe in order to draw the nail-wall away from the nail. I am unable to say how effective these operations are, but there seems no justification for making a scar at the side of the toe when a smaller scar on the dorsum will suffice.

There is another group of operations for embedded nail which is designed to remove parts of both the nail and nail-wall. A typical example is the wedge excision advocated by Watson-Cheyne and Burghard (1912) and repeatedly described with various modifications since then (O'Donoghue, 1940; Hyde, 1945; Lake, 1952). A wedge of tissue consisting of nail-wall, a strip of nail, and the underlying matrix is excised and the wound either left open or partly or completely closed with sutures (*Fig.* 440). Although wedge excision is a standard operation described in most text-books of surgery it is neither logical nor efficient. It is illogical because it is unnecessary to remove both the factors in a disorder due to two incompatible factors; if the nail is eliminated then the sepsis will always disappear. Wedge excision is inefficient because the very structure which it is most important to remove is least well exposed. *Fig.* 440 is copied from a description of the operation and shows that the wedge removed is so shaped that at the proximal end and at the depth of the matrix the removal of tissue is minimal. It is an operation which promotes drainage rather than excision of the germinal matrix.

Disillusioned by the frequent recurrences following toe-nail operations many surgeons have resorted to operations which are designed to eliminate the whole nail complex. This was the treatment originally employed by Dupuytren (1826), who left a large raw area to granulate. The problem of skin-cover has been solved by skin-grafting (Dolan, 1935), by advancing a dorsal skin-flap (Nuttall, 1941), and by partial or complete amputation of the terminal phalanx.

The last alternative, sometimes called the "terminal Syme" amputation, is frequently performed to-day (*British Surgical Practice*, 1948), but it is altogether disproportionate to the purpose for which it is employed—an interference with the skeleton in order to remove a tiny piece of epidermis. It is claimed that terminal amputation is a certain cure, but, ironically enough, it is not. The proximal limits of the germinal matrix may be left behind and I have seen 3 cases where nail-tissue has sprouted through the suture line (*Fig.* 441). Even if the cure is complete it remains a mutilating operation which is only justified when there are additional reasons for shortening the toe, such as arthritis or deformity of the toe.

SUMMARY

1. Growth of nail, whether normal, embedded, or onychogryphotic, only takes place from the germinal matrix. This is confined to the lunular area of the nail-bed.

2. Techniques for total and partial excision of the germinal matrix are described. These are based on operations described by Quénu in 1887.

3. One hundred results are reported; there was one failure due to regeneration of nail.

4. It is concluded that, with a few exceptions, excision of the germinal matrix is a satisfactory treatment for surgical disorders of the nail.

I wish to acknowledge the co-operation of many surgeons who have allowed me to operate on their patients. I am indebted to Mr. A. L. Eyre-Brook and Mr. J. G. H. James for help in revision of the paper. I am grateful to Mr. K. H. Pridie for *Fig.* 441. *Fig.* 431 is reproduced by courtesy of Sir W. Le Gros Clark and the Clarendon Press, Oxford.

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SHORT NOTES OF RARE OR OBSCURE CASES

A CASE OF OLEOGRANULOMA OF THE RECTUM

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An oleogranuloma is the resulting inflammatory tumour stimulated by the presence of oil, wax, or paraffin in the tissues. Only certain individuals, possibly because of allergy, produce an exaggerated reaction to the oil (used from now on to indicate also wax or paraffin). It appears that the oil, or derivatives of it, stimulate a surrounding granulomatous inflammation characterized by the presence of an increased number of lymphocytes, foreign-body multinucleate giant cells, and fibroblasts. Some of the oil particles appear to pass along lymphatics to the lymph-nodes, thus resembling pathologically and clinically a neoplastic or bacterial spread; others are dispersed locally by the effect of gravity and muscle contraction. Thus from a localized deposit of oil a spreading inflammatory lesion occurs, finally resulting in some cases in a dense hyalinized fibrosis. Macroscopically the lesion usually presents a yellowish hue when cut across.

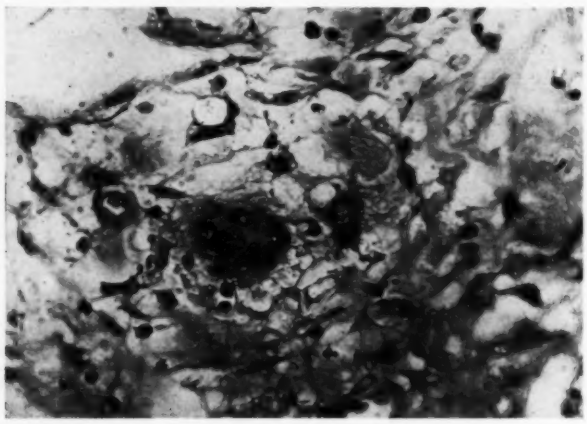

FIG. 442.—Shows typical multinucleate giant cells, macrophages, and lymphocytes. (× 278.)

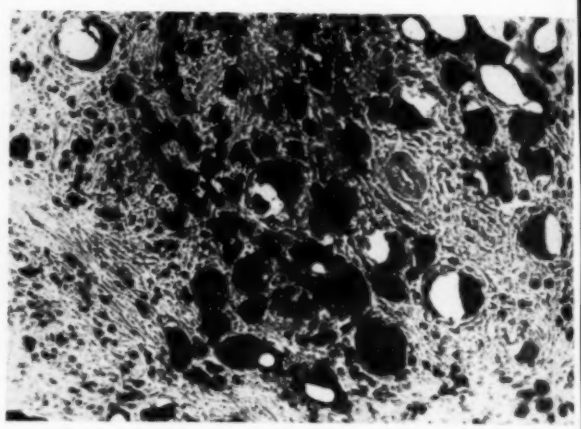

FIG. 443.—First stained with osmic acid which is taken up by human fat (appears black), then with Scharlach Red which is taken up by vegetable oil (almond oil) and appears somewhat paler in this type of photograph. (× 70.)

Not all oils are equally tumefactive. It is thought that mineral oils are most so, vegetable oils least so, while animal fats occupy an intermediate position. The fatty acids and their calcium salts resulting from the breakdown of human fat appear to stimulate the granulomatous tumour of traumatic fat necrosis.

The oil which stimulates the formation of an oleogranuloma may gain entry to the tissues by deliberate injection, as with wax for cosmetic reasons (Andrews, 1954) or with phenol in almond oil for hæmorrhoids (Jackman, 1940); accidental inoculation may occur with those using high-pressure grease guns (Moore, 1946). In other cases repeated contact of the oil with an epithelial surface such as brilliantine on the scalp (Robb-Smith, 1948), or liquid paraffin ingested as an aperient (Jaffé, 1934) have resulted in granulomata of scalp and appendix respectively. The insertion of a petroleum-jelly gauze pack into an ischiorectal abscess cavity (Symmers, 1955) and liquid paraffin into the peritoneal cavity to prevent adhesions (Cruickshank, 1941), as well as iodized oils used in radiography for bronchograms, salpingograms, and myelograms, have all resulted in granulomatous lesions.

The following is a brief description of an oleogranuloma occurring in the rectum.

CASE REPORT

Mrs. C. E., aged 54 years, reported to hospital on May 8, 1956 complaining of pruritis ani and prolapse of hæmorrhoids. Examination revealed second-degree hæmorrhoids. Sigmoidoscopy was normal to 22 cm. An injection of 3 c.c. of 5 per cent phenol in almond oil was given into each primary hæmorrhoid at the level of the anorectal ring.

The patient was subsequently seen at monthly intervals for the next three months and received further injections to a total of 23 c.c. of phenol in almond oil.

Three months after the first injection she complained of tenesmus and low backache, with slight discharge noted on her underclothes. No local abnormality was noted in the rectum at the time. One month later a firm irregular swelling over the front of the lower sacrum was felt. It was fixed to the periosteum but the mucosa over it moved easily and appeared normal on sigmoidoscopy. X-ray examination of the pelvis and sacrum was normal. In view of the doubtful nature of the lesion, surgical exploration through a transverse incision at the level of the sacrococcygeal junction was carried out. After excising the coccyx the fascia of Waldeyer was opened.

The swelling was found to be fused both to the periosteum of the sacrum and to the fascia propria of the rectum. As much as possible of the abnormal tissue was excised and 'frozen section' revealed it to be of an inflammatory nature. Later, microscopy of paraffin-block sections showed an oleogranuloma with typical cells around deposits of almond oil (*Figs.* 442, 443).

The patient made a satisfactory recovery and has had no further symptoms or signs.

COMMENT

The clinical similarity between an oleogranuloma and a malignant neoplasm may be such that only biopsy and microscopical examination will confirm one or other. It is important to be on guard where a history of previous oil insertion is elicited at the site of a tumour. This may prevent a mutilating operation for a non-malignant condition. In 6 cases of oleogranuloma described by Symmers (1955), 2 had an abdominoperineal excision of rectum, 1 a radical mastectomy, and 1 a bilateral orchidectomy. The appearance of a granulomatous tumour after the insertion of oil may take weeks, months, or years depending on the individual's particular response and the volume of oil inserted (Jackman, 1940). The most acute phase of the inflammatory reaction is early and in the rectum may cause tenesmus and low backache. The resulting lesion in the rectum may be either a nodular tumour or an annular stricture, its position depending on the site of injection (Jackman).

The mucosa over the tumour is commonly mobile, occasionally fixed, and scarred. Differentiation from a carcinoma, sarcoma, chordoma, teratoma, fibromyoma, subacute inflammatory anorectal lesions, and lymphogranuloma inguinale must be made.

Malignant change in an oleogranuloma is doubtful, but Goldenberg (1954) records 2 cases of fibrosarcoma of the buttock occurring at the site of previous injections of penicillin in sesame oil. He makes no mention, however, of oil particles among the tumour cells in his microscopical descriptions of the tumours.

Jackman (1940) recommends hot retention enemas and anti-constipation diet as the treatment of the anorectal oleogranulomatous tumours and strictures. In his opinion forcible dilatation and proctotomy give only temporary relief.

As a general principle, if practical, an early and widespread removal of the oil by incision, excision, or both should be carried out.

I wish to thank Mr. A. E. Williams, Consultant Surgeon, Hampstead General Hospital, for permission to publish this case.

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INTRAMURAL HÆMATOMA OF THE DUODENUM

A REPORT OF A CASE AND A REVIEW OF THE LITERATURE

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A HÆMATOMA of the duodenum or upper jejunum, presenting as a tense dark blue upper abdominal mass, is a rare and vivid finding at laparotomy or autopsy. Since its first description by Pilliet in 1895, as reported by Tempsky (1925), we have been able to trace only a few reports of this phenomenon in the literature, to which the following case is added.

CASE REPORT

B. G., a boy aged 6 years, was admitted to the Children's Hospital, Sheffield, on Aug. 1, 1954. He gave a history of 36 hours' central aching abdominal pain accompanied by frequent bilious vomiting. Examination revealed an afebrile, healthy, but rather dehydrated child who was obviously in considerable pain. The abdomen was generally tender, particularly in its upper part, where muscle guarding was found. Careful palpation revealed a tender rounded swelling, about 2 in. in diameter, projecting from below the costal margin just to the left of the midline. The white-cell count was 17,000 and the urine normal.

It was thought that this must be a case of intussusception occurring in a somewhat older child, and operation was carried out shortly after admission, his condition having been improved by intravenous fluid. Laparotomy revealed a mass in the left upper quadrant of the abdomen involving the distal half of the duodenum and the duodenojejunal junction. This part of the bowel was grossly enlarged, dusky purple in colour, and clearly obstructing the lumen of the bowel. This area was clearly demarcated from healthy bowel above and below (*Fig.* 444).

It was decided to perform a gastro-enterostomy in order to by-pass the obstruction. When the first loop of the jejunum was opened, it was found that distended purple mucosa projected distally for a distance of 1–2 cm. within the lumen of healthy bowel. It was evident that the mucosa had been stripped by hæmatoma from the underlying layers and completely occluded the bowel lumen. An isoperistaltic retrocolic gastro-enterostomy was performed. At the completion of the anastomosis an incision was made into the discoloured wall of the duodenojejunal junction. Some dark altered blood was sucked out, allowing the wall to collapse a little, but no attempt was made to evacuate the hæmatoma completely. The post-operative course was quite smooth and the child was discharged home at the end of 3 weeks.

The parents were again questioned after operation and the information was then obtained that the pain had come on immediately after the patient had received a blow on the abdomen whilst playing with some other children.

Table I.—DETAILS OF 12 CASES OF DUODENAL HÆMATOMA

CASE	AUTHOR	SEX	AGE	HISTORY	PHYSICAL SIGNS	CLINICAL DIAGNOSIS	TREATMENT	RESULTS	NOTES
1	*Non-traumatic Origin* Oppenheimer (1933)	F.	42	4 d. pain and vomiting	Vague mass in left upper abdomen	Acute pancreatitis	Laparotomy and drainage	Death	Carcinoma of head of pancreas causing an acute pancreatitis presumably with erosion of a vessel in the duodenal wall
2	Glass (1937)	?	Newborn	Vomiting of blood, jaundice	Bruises and petechiæ. Palpable liver and spleen	Bleeding diathesis	Blood transfusions	Death	
3	*Idiopathic Origin* Dey (1952)	F.	6	2 d. pain and bilious vomiting	Hard tender mass to right of the umbilicus		Laparotomy, evacuation of hæmatoma in second part of duodenum	Recovery	No history of trauma, no bleeding diathesis
4	Case of Brannan's reported by Felson and Levin (1954)	M.	33	2 d. hiccough. No abdominal pain	Upper abdominal guarding		Laparotomy, evacuation of hæmatoma 7 cm. diameter in third part of duodenum	Recovery	No history of trauma
5	*Traumatic Origin* Pilliet, quoted by Tempsky (1925)	?	?	?	?	?	?	Death	Post-mortem report
6	Liverud (1948)	M.	3	2 d. pain and bilious vomiting	Abdomen soft. No masses felt	Benign tumour of jejunum (following barium meal)	Laparotomy, hæmatoma 10 cm. distal to duodenojejunal flexure. Gastro-enterostomy	Recovery	
7	Stirk (1953)	M.	9	3 d. pain and vomiting ('coffee ground')	Abdomen distended. Guarding. Ill-defined upper abdominal mass		Laparotomy, hæmatoma second part of duodenum to proximal 4 in. of jejunum evacuated	Recovery	
8	Cooling (1953)	M.	6	4 d. pain and bilious vomiting	Abdomen rigid	Ruptured spleen	Laparotomy, hæmatoma 6 in. long in upper jejunum thought to be gangrenous and resected	Recovery	
9	Felson and Levin (1954)	M.	18	4 hr. pain and vomiting	Upper abdominal tenderness	Traumatic pancreatitis, ? ruptured duodenum	Laparotomy, evacuation of hæmatoma in distal duodenum and proximal jejunum	Recovery	
10	Felson and Levin (1954)	F.	9	2 d. upper abdominal pain, vomiting	Abdominal tenderness and guarding	Duodenal hæmatoma. Diagnosed on barium meal	Conservative	Recovery	Typical appearance on barium meal of hæmatoma of third part of duodenum
11	Case of Fry's reported by Felson and Levin (1954)	M.	9	6 hr. abdominal pain and nausea	Diffuse abdominal tenderness	1. Acute appendicitis. 2. Duodenal polyp (after barium meal)	1. Appendicectomy. Ninth day laparotomy and evacuation of hæmatoma in third part of duodenum	Recovery	History of trauma not obtained until after operation
12	Present authors	M.	6	36 hr. central abdominal pain and bilious vomiting	Upper abdominal guarding, mass in left upper quadrant	Intussusception	Laparotomy, hæmatoma of distal half of duodenum and duodenojejunal flexure. Gastro-enterostomy	Recovery	History of trauma not obtained until after operation

A barium meal carried out 2 months post-operatively revealed a normal stomach and duodenum with no obstruction; the gastro-enterostomy was not functioning.

DISCUSSION

Aetiology.—The relevant details of this patient and the 11 other published reports are presented in *Table I*.

In 2 patients the duodenal hæmatoma was an incidental finding secondary to other pathological disturbances (in one a bleeding diathesis, and in

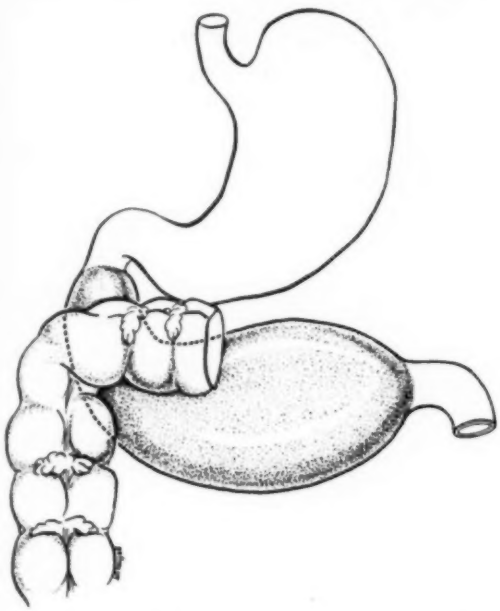

FIG. 444.—Showing enlarged area of the bowel.

the other a pancreatitis complicating a carcinoma of the head of the pancreas).

In the remaining patients the hæmatoma was the primary lesion. Eight had a definite history of trauma up to 48 hours preceding the onset of symptoms, but in the remaining 2 patients (one a child and one an adult) no such history could be obtained, although the operative findings closely resembled the traumatic cases.

Clinical Features.—Of particular interest is the fact that 7 of the patients with traumatic or idiopathic hæmatomata were children between the ages of 3 and 9, a surprising contrast to other intra-abdominal closed injuries, which predominantly occur in young adults.

Upper abdominal pain and frequent bilious vomiting were characteristic symptoms, but a palpable mass was found in only 3 cases including our own; this in spite of the fact that in only one patient was abdominal rigidity (as opposed to tenderness and guarding) noted.

Radiology.—A plain abdominal radiograph in Stirk's case (1953) revealed a large amount of gas in the stomach. Felson and Levin (1954) describe an obscured or obliterated psoas shadow on the right side.

A barium meal was carried out in the patient reported by Liverud (1948) and here a diagnosis of a benign tumour of the jejunum was made pre-operatively in the absence of a history of trauma. Felson and Levin give a careful description of the barium meal findings in their 4 cases and note, together with Liverud, the smooth, sharply defined intramural mass and the crowding together of the valvulæ conniventes producing the coiled-spring effect more familiarly seen in intussusception. This characteristic appearance enabled them to make a confident diagnosis in one patient who was subsequently treated conservatively, with recovery.

Comment.—The duodenum and duodeno-jejunal junction are particularly liable to injury in closed abdominal injuries because of the relatively fixed position and relationship to the vertebral column. Rupture of the bowel at this site produces vivid symptoms leading almost invariably to laparotomy. It is possible that a hæmatoma of the duodenum is of relatively frequent occurrence, but unless it reaches sufficient size to obstruct the bowel lumen it may not produce symptoms which occasion alarm and lead to laparotomy.

When bilious vomiting and upper abdominal pain follow a closed injury, but where rupture of a viscus can be excluded on the clinical signs, a barium meal examination may enable a diagnosis of an intramural hæmatoma to be made and the patient treated by conservative measures with some confidence.

The possibility of trauma should be borne in mind in every obscure abdominal emergency in childhood, since a history of injury may be difficult or even impossible to obtain.

SUMMARY

1. A case of traumatic hæmatoma of the duodenum in a child of 6 is reported.

2. The literature on the subject of duodenal hæmatoma is reviewed.

Acknowledgement.—We would like to thank Mr. R. B. Zachary for his permission to publish this report of the patient upon whom he operated, as well as for his advice in the preparation of the paper.

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ERRATUM

Andrew M. Desmond and Paul W. Seargeant: "The Place of Primary Gastric Resection in the Treatment of Perforated Peptic Ulcer," 1957, Vol. XLV, No. 191, p. 284.

For 2. With perforated *gastric* ulcer over 40 years.

read 2. With perforated *duodenal* ulcer over 40 years.

URETERIC SECONDARIES FROM A HYPERNEPHROMA

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THIS brief communication contains a report of secondary deposition in the ureteric wall from a hypernephroma of the kidney and refers to other cases described in the literature.

FIG. 445.—Right retrograde pyelogram.

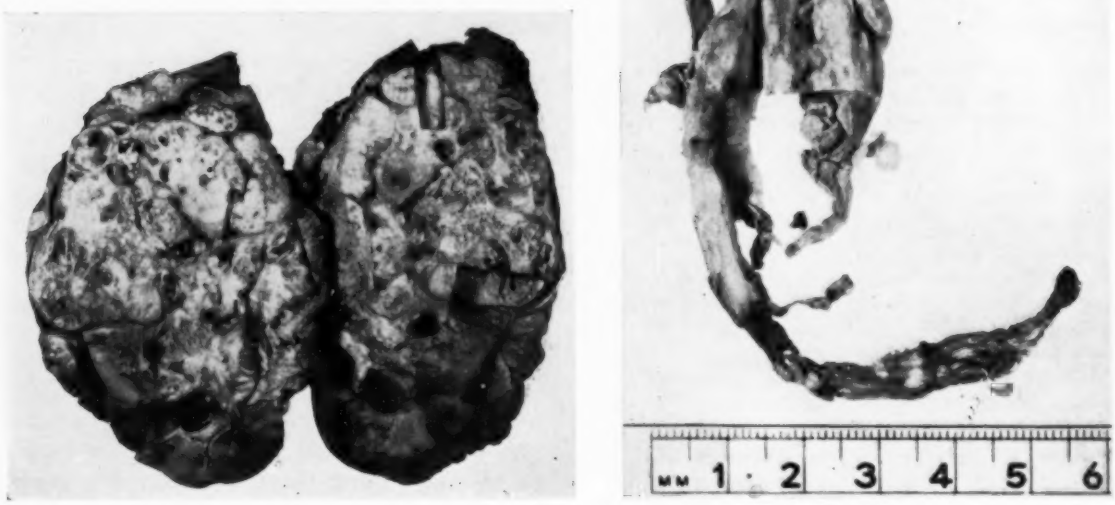
CASE REPORT

Mr. C. I., aged 53, a lorry driver, first attended Hammersmith Hospital in May, 1956. He complained of painless hæmaturia for the preceding 2 days, but had no other urinary symptoms. He had no loin pain.

Physical examination revealed some enlargement of the right kidney and a midstream specimen of urine showed red blood-cells. Cystoscopic examination was normal, but when repeated one week later after the patient complained of pain in the right loin, blood-clot was seen extruding from the right ureteric orifice.

Excretion pyelograms showed an enlarged right kidney with poor concentration of the radio-opaque material. Right retrograde pyelography was performed and was accompanied by brisk hæmorrhage from the right ureter. The pyelograms (*Fig.* 445) showed an enlarged right kidney with gross deformity of the pelvis and upper ureter and cavitation of the superior calices. Some filling defects could be seen in the renal pelvis and calices. Filling defects were also seen in the ureter approximately 1 in. distal to the pelvi-ureteric junction. Clot and debris collected from the right ureteric catheter were examined for malignant cells, but none was seen.

The pre-operative diagnosis was carcinoma of the renal pelvis with ureteric seedlings, and operation was advised. The right kidney was exposed through a Morris's muscle-cutting loin incision and was found to be 6 × 4 in. in size, firm and adherent to the perinephric fat. Dissection of the renal pedicle revealed two renal veins, which were ligated and divided flush with the vena cava;



A B

FIG. 446.—Kidney split open to show (A) tumour in upper pole and pelvis, (B) norma lintervening ureter and mammillated tumorous ureteric mucosa. (× ⅔.)

the lower renal vein contained tumour for 1 in. of its length. The renal pedicle was divided, the pelvis and ureter were mobilized but not divided, and the kidney was brought out on to the loin. The loin incision was closed, the lower part of the ureter was exposed through a right gridiron extraperitoneal incision, and the kidney with the attached ureter was avulsed from the loin. The patient's post-operative course was uneventful.

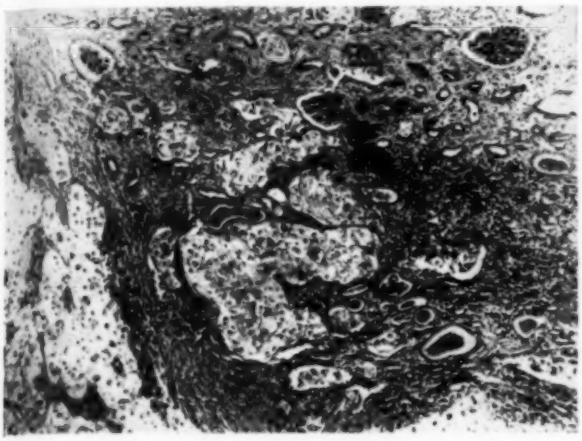


FIG. 447.—Kidney showing tumour edge growing into surrounding normal kidney. (× 45.)

Macroscopic Findings (*Fig.* 446).—Nearly all the kidney was replaced by friable tumour, which was also present in the mucosa of the pelvis and in the ureter at a site about 1 in. below the pelvi-ureteric junction. The intervening uppermost part of the ureteric mucosa was intact. A large vein at the hilum contained tumour.

Microscopic Findings.—Sections taken from the kidney (*Fig.* 447), the intervening apparently normal ureter (*Fig.* 448), and the attached portion of the ureter (*Fig.* 449) were all found to contain hypernephromatous tissue. That part of the ureter in which the mucosa was free from tumour contained several intramural veins filled with tumour.

Fig. 448 shows tumour in ureteric veins; *Fig.* 449 shows tumour growing more closely to the ureteric lumen, with the mucosa, although thinned out over the tumour, still virtually intact.

The patient was given a full course of radiotherapy to the loin, but a subsequent chest radiograph showed a left lower lobe solitary metastasis, and he was referred to the thoracic surgeon for consideration for lobectomy. Rapid development of numerous pulmonary metastases, however, contra-indicated thoracotomy.

DISCUSSION

The pathological findings leave no doubt that this was a case of hypernephroma with deposits in the wall of the ureter. Macroscopically the intervening ureteric mucosa was free from deposits, but microscopy showed the presence of tumour in the ureteric veins. Since the lower renal vein was filled with tumour it is postulated that the method of ureteric involvement was retrograde spread from the renal vein down a ureteric vein.

That ureteric spread from a hypernephroma is rare is borne out by the absence of any reference to the phenomenon in the extensive review of renal and ureteric tumours by Riches, Griffiths, and Thackray (1951).

Willis (1953) quotes a single case reported by MacAlpine (1948), with metastases in the ureteric stump which became overt five years after nephrectomy for hypernephroma. MacAlpine considered this to represent mucosal implantation via the lumen of the ureter, and did not comment on the apparently normal intervening mucosa or on the state of the ureteric veins.

FIG. 448.—Apparently normal ureter containing tumour in the ureteric veins. (× 22·5.)

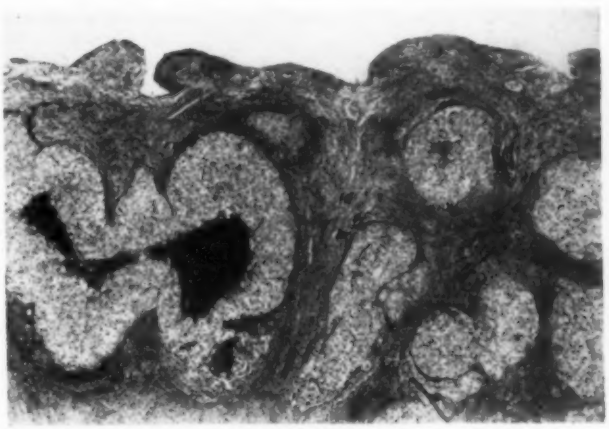


FIG. 449.—Section taken at level of macroscopic ureteric tumour showing hypernephromatous tissue growing closely to the lumen but covered by intact mucosa. (× 22·5.)

Retrograde venous spread of hypernephroma to the vagina has been recognized (Martzloff and Manlove, 1949) and it is possible that a similar method of spread accounts for some of the deposits found in the suprarenal (Riches and others describe 34 such depositions out of 562 cases of hypernephromatous metastases, but make no reference to this possibility). There can be little doubt about the mechanism of spread in the present case and it is possible that the same process occurred in the case described by MacAlpine. It is possible also that the rare cases of hypernephromatous metastases to the bladder (Hoffman, 1907) have a similar aetiology.

SUMMARY

A rare type of venous spread to the ureter in a case of hypernephroma is recorded and the few

references in the literature to ureteric involvement by such tumours are quoted.

Acknowledgements.—I am indebted to Dr. A. G. E. Pearse, Department of Pathology, for the histological reports on this case. My thanks are due to Mr. R. Shackman and Professor Ian Aird for permission to publish this case, and to Miss R. Klein for the photographs.

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RELAPSING PANCREATITIS

REPORT OF A CASE WITH UNUSUAL FEATURES

BY R. B. WRIGHT
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CHRONIC relapsing pancreatitis is not a common condition and few individual clinicians have a wide experience of it. Thus Cattell and Warren (1953) of the Lahey Clinic report on a series of 38 cases. They point out that chronic relapsing pancreatitis is to be regarded as " a progression of acute pancreatitis with important additional factors which ensure recurrent episodes and ultimately lead to permanent disease attended by chronic symptoms and physiologic disturbances ". Various ' additional factors ', e.g., alcohol, trauma, disease of the biliary tract, infection, and anatomical variants of the duct terminations (Doubilet and Mulholland, 1949), have been suggested without any very convincing proof. The present case is reported because a more tangible anatomical lesion was demonstrated which may have had an important bearing on the aetiology of symptoms.

CASE REPORT

The patient, a well built male aged 41, with no previous dyspepsia was first admitted to hospital on April 1, 1952, in obvious pain, grey and shocked. Severe epigastric pain had come on very suddenly two hours previously ; he had vomited once, there was no radiation of pain beyond the abdomen, which was rigid and tender all over, and silent. A diagnosis of perforated peptic ulcer was made and laparotomy performed one hour after admission. There was no free fluid in the peritoneal cavity ; the stomach, duodenum, and gall-bladder appeared normal, but the pancreas was noted to be grossly swollen and firm ; there was no fat necrosis. A diagnosis of acute pancreatitis was made and the abdomen closed. The patient made a smooth recovery and was discharged on April 11. On May 8 he was re-admitted with severe epigastric pain and suffering from shock. Morphine gr. ¼ was given by intravenous injection and within twenty-four hours he had recovered. On this occasion the urinary diastase was

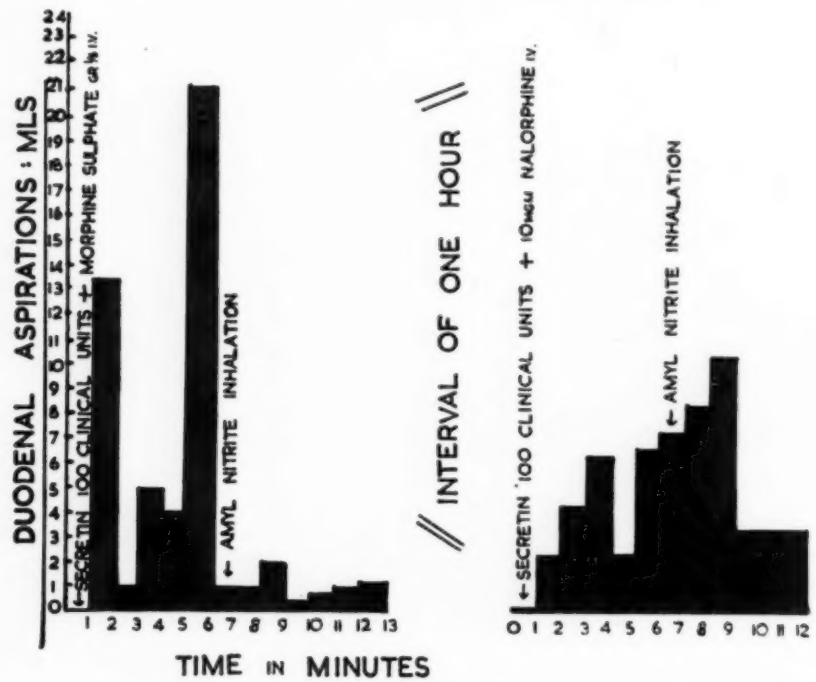
DUODENAL ASPIRATIONS : MLS
← SECRETIN 100 CLINICAL UNITS + MORPHINE SULPHATE gr ¼ IV.
← AMYL NITRITE INHALATION
// INTERVAL OF ONE HOUR //
← SECRETIN 100 CLINICAL UNITS + 10mgm NALORPHINE IV.
← AMYL NITRITE INHALATION
TIME IN MINUTES

FIG. 450.—Secretin test.

200 units. Three months later the patient had another similar attack of severe pain accompanied by diarrhœa. He continued to have similar but less severe attacks at varying intervals until May 7, 1953, when he was again admitted to hospital with a very severe attack; on this occasion his serum diastase was 500 Wohlegemuth units. Morphine again relieved his symptoms. A cholecystogram showed no abnormality; barium meal showed some spasm of the duodenal cap. The glucose tolerance curve was within normal limits; there was no steatorrhœa.

FIG. 451.—Mucosal polyp, stained. H. and E. (× 35.)

Thereafter, the patient continued to have repeated attacks of severe pain, preceded by a curious aura of warmth and tingling in the upper limbs. He found that he obtained most relief by adopting the knee-elbow position. The pain was so severe and the aura so alarming that he always carried an omnopon tablet with him to his work. He would take the tablet and leave his work bench as soon as the aura began and adopt the knee-elbow position in anticipation of the pain. His wife noted that he was becoming very irritable and anxious and constantly haunted by the dread of another attack of pain. Towards the end of 1956 the pain had become so frequent and severe that the patient begged for surgical relief. He was admitted to hospital on Nov. 24 and the same night had a very acute attack in which his serum amylase rose to 400 units and his urinary diastase to 1600 units. A biligrafin cholecystogram showed an apparently normal gall-bladder and bile-duct system; a secretin test (*Fig.* 450) showed no evidence of sphincter blockage. At laparotomy the pancreas was swollen, rounded, and firm but there was no evidence of calcification; all other abdominal viscera appeared normal. The duodenum was opened and the papilla of Vater inspected; it appeared normal, but when a bougie was passed down the common bile-duct a small congested pedunculated polyp (*Fig.* 451) was pushed through the papilla. This seemed a possible cause of intermittent blockage of the ampulla and it was therefore excised and sphincterotomy performed. The patient made a good recovery and has had no further attacks of pain; he has lost his anxious look, regained lost weight, and also his enjoyment of food and life.

DISCUSSION

There is no rational treatment for chronic relapsing pancreatitis and the procedures recommended include excision of the pancreas—partial or total pancreatic resection, vagotomy, cholecystectomy, choledochostomy, sphincterotomy, and gastrojejunostomy. No procedure is uniformly successful and many patients have had several operations. There is seldom any convincing indication of which procedure is likely to be successful. It is much too soon to make any claim for the value of removal of the polyp in this particular case, but it does seem possible that intermittent impaction of the polyp in the sphincter of Oddi was the underlying cause of pancreatic obstruction. It is suggested that downward bouginage of the common bile-duct is a worth-while procedure before sphincterotomy is performed.

SUMMARY

1. A case of relapsing pancreatitis is reported in which a polyp, capable of blocking the sphincter of Oddi, was demonstrated.

2. The condition was relieved by removal of the polyp.

3. The patient had a well marked prodromal aura and found some relief by posture.

REFERENCES

CATTELL, R. B., and WARREN, K. H. (1953), *Surgery of the Pancreas*. Philadelphia: W. B. Saunders.

DOUBILET, H., and MULHOLLAND, J. H. (1949), *Surg. Clin. N. Amer.*, **29**, 333.

REVIEWS AND NOTICES OF BOOKS

The Collected Papers of Hugh Trumble. Edited by LEONARD COX, M.D., R. S. LAWSON, F.R.C.S., and T. E. LOWE, M.D. Pp. 302 + xxi. 1957. Alfred Hospital, Melbourne, in association with Melbourne University Press. 84s.

IT is given to few surgeons to have their surgical writings republished in book form by their colleagues.

These collected papers of Hugh Trumble of Melbourne cover many branches of surgery and each individual paper has something to tell the reader. Mr. Trumble's surgical background has been unique, for he has been able to practise general surgery in the true sense and cover such branches as orthopædic surgery and neurosurgery. His extra-articular arthrodesis of the hip-joint has been adopted throughout the ever-expanding orthopædic world. As a neurosurgeon Trumble excelled and was the pioneer who placed this modern branch of surgery in its proper place in Australia.

This volume, which surgeons both at home and abroad should read, is beautifully produced and the editors are to be congratulated on their fine work. Several of the articles have been reprinted from the BRITISH JOURNAL OF SURGERY.

The Forequarter Amputation. By H. F. MOSELEY, M.A., D.M., M.Ch., F.R.C.S., F.A.C.S., Hunterian Professor, Royal College of Surgeons of England; Assistant Professor of Surgery, McGill University, Associate Surgeon, Royal Victoria Hospital, Montreal, Canada. 11 × 8¾ in. Pp. 79 + viii, with 25 illustrations and 11 colour plates. 1957. Edinburgh and London: E. & S. Livingstone Ltd. 42s.

THIS is a most useful monograph which comprises a complete description of this formidable operation and contains a wealth of illustrations depicting the various stages of the procedure. Mr. Moseley has already written a monograph of *Shoulder Lesions* and this further work will be of real value to any surgeon who is called upon to perform this difficult exercise in surgical technique.

The author gives an interesting account of the early cases reported by Paul Berger, Buchanan, and Fowler. Various illustrative cases are reported and cases of scapulectomy are included.

An excellent list of general references is incorporated in this monograph.

As a reference volume this work could not be bettered.

Clinical Proctology. By J. PEERMAN NESSELROD, B.S., M.S., M.Sc., M.D., F.A.C.S., F.A.P.S., Assistant Professor of Surgery, Northwestern University Medical School; Attending Surgeon, Division of Proctology, Evanson Hospital, Evanson, Illinois. Second edition. 9½ × 6 in. Pp. 296 + xx, with 72 illustrations. 1957. Philadelphia and London: W. B. Saunders Company Ltd. 49s.

AN excellent feature of this, the second edition of Dr. J. P. Nesselrod's book on clinical proctology, is the emphasis which he lays on meticulous care and accuracy in diagnosis. All the routine steps are described in great detail, and it is refreshing to note that the author advocates simple as opposed to elaborate instruments and techniques. One has to get used to the idiom that some of the operation drawings are with the patient in the jack-knife position, but the essential quadrants are labelled and therefore easily identified. The author is a firm believer in the principle of external drainage of anal wounds, and this is exemplified in the lateral drainage wounds depicted after amputation of a partial prolapse. The treatment of fistula is set out in a very sensible and not over-elaborated way, and it is clear that the author practises and recommends the operation of 'fistulotomy', which corresponds to the old well-established method of 'lay open' as practised in this country since the days of Frederick Salmon and William Allingham. Major surgery is not described except for its indications, and good references are given to recommended procedures. The author is clearly very interested in polyps and all aspects are discussed in detail. The advantages of coloscopy either at the time of a laparotomy and colotomy, or via a colostomy stoma, are indicated, and many practical details about fulguration of polyps are given.

The usual importance which one expects in American text-books is given to the anal crypts and the operation of 'multiple cryptotomy' is illustrated: this appears to be a very radical procedure and one may be allowed to wonder if it is really necessary and how much scar tissue is left in the anal canal and at the anal margin from these multiple incisions. The author is unenthusiastic as regards the merits of injection therapy for hæmorrhoids and fissures, and he is opposed to office surgery.

The coloured sigmoidoscopic illustrations are an interesting feature of this book and were prepared by a photographic process for which the author and his associates are well known in America.

The Foreword is written by Dr. Louis A. Buie, whose influence in regard to teaching and methods is easily seen throughout the book and frequently referred to. Within the small compass of less than 300 pages the author gives a carefully considered, balanced, and practical account of proctology which is well worthy of study by all who are interested in this subject. The work is, we feel, eminently sound and gives the impression throughout of the author's care for the patient's welfare. The relationship of proctology to medicine generally, whether it be pædiatrics, geriatrics, gynæcology, or urology, is discussed in considerable and interesting detail, and the author rightly advises anyone to "relegate to a qualified colleague any service which he feels he cannot perform creditably himself". This is indeed sound and proper advice to appear in a book on a special subject such as this; when acted upon it certainly leads to improved team-work and co-operation and it is advice which we can wholeheartedly endorse.

The Surgical Management of Pulmonary Tuberculosis. Edited by JOHN D. STEELE, M.D., with a Biographical Sketch of John Alexander, by CAMERON HAIGHT, M.D. 10¼ × 7¼ in. Pp. 213 + xxviii, with numerous illustrations. 1957. Springfield, Illinois: Charles C. Thomas (Oxford: Blackwell Scientific Publications). 72s.

THE pupils of the late John Alexander, the famous pioneer in thoracic surgery, pay a graceful tribute to his memory in this volume, which is the first of a series on the various phases of thoracic surgery. Fifteen collaborators have produced a book of some 200 pages which provides a succinct but delightfully lucid account of the present position of thoracic surgery in the treatment of lung tuberculosis.

With the complete change in outlook that has followed the full use of chemotherapy, and of streptomycin, it is most valuable to have a volume which includes a clear description of the value of surgery in modern practice. There is an especially valuable chapter on chemotherapy in tuberculosis, which covers the subject adequately for

surgeons. The indications for excision and collapse therapy are presented in a balanced manner and this work will be most helpful to physicians and surgeons concerned in the management of pulmonary tuberculosis.

Fractures and Related Injuries. By J. GRANT BONNIN, M.B., B.S. (Melbourne), F.R.C.S. (Eng.), Consultant Orthopædic Surgeon, Central Middlesex Hospital; late Orthopædic Adviser, South-East Asia Command; Hunterian Professor, Royal College of Surgeons. 9 × 6 in. Pp. 710 + x, with 775 illustrations. 1957. London: William Heinemann (Medical Books) Ltd. 84s.

THIS is a larger edition of the author's previous volume, *A Complete Outline of Fractures*. As such it is no longer a practical proposition for the undergraduate to use, but may prove valuable to the newly qualified practitioner who is confronted with the everyday management of fractures in a general hospital. The whole subject is dealt with very comprehensively, but there is little new in the book, and apart from the excellent chapter on ankle injuries, it has little to offer to the practising orthopædic or accident surgeon. Although a full account of the treatment of each injury is given, the reader is sometimes left in a little confusion as to what is the best thing to do for a particular condition. For instance, one might be left with the impression that the author favoured the manipulative reduction of a fracture-dislocation of the cervical spine over the slower and much safer reduction by continuous traction. In a similar way, perhaps too much emphasis has been laid on the value of treating simple fractures of the upper end of the humerus on abduction frames and the like, when it is realized that most fracture surgeons to-day would consider such treatment as unnecessary. The X-ray reproductions are not of the highest order and it is difficult on many occasions to appreciate points which have been made in the text. In contradistinction, however, the diagrams and illustrations of apparatus are excellent, and for these alone the book should be a very useful reference for anyone who is not conversant with some of the complicated pieces of 'machinery' used in fracture surgery.

Methods in Surgical Pathology. By HENRY A. TELCH, M.D., Assistant Professor of Pathology, Northwestern University Medical School, Chicago. 9 × 5¾ in. Pp. 127 + viii. 1957. Springfield, Ill.: Charles C. Thomas (Oxford: Blackwell Scientific Publications). 36s.

THIS monograph is written primarily for the resident pathologist who has to deal with surgical pathological material. The various techniques in use for the processing of material removed in the operating theatre are set out in detail. The preparation of paraffin blocks and the staining of sections are described in an interesting and practical way.

An appendix includes sample protocols of gross descriptions of representative surgical specimens.

A useful book for the young pathologist.

Practical Otolaryngology. By GERVAIS WARD MCAULIFFE, M.D., F.A.C.S., F.I.C.S., Associate Clinical Professor of Otolaryngology, Cornell University Medical College. 8 × 5 in. Pp. 320, with 13 illustrations. 1957. New York: McGraw-Hill Publishing Co. Ltd. 52s. 6d.

THIS is a small practical guide to the study of common diseases of the ear, nose, and throat. It is intended as a handbook for the general practitioner who wishes to know the practical side of otolaryngology. It should also prove of value to senior students who wish to have a practical book to guide them in this important subject.

Tumour Surgery of the Head and Neck. By ROBERT S. POLLACK, M.D., F.A.C.S., Clinical Instructor in Surgery, Stanford University School of Medicine. 10 × 6½ in. Pp. 101, with 49 illustrations. 1957. London: Henry Kimpton. 37s. 6d.

THIS monograph, which is written in an interesting and lucid manner, should appeal not only to the general surgeon but also to the plastic surgeon and those laryngologists who are interested in malignant disease of the neck. The illustrations throughout the book are mostly black and white drawings and are quite original, which is refreshing in these days when standard illustrations seem to be copied from one book to another.

The book deals for the most part with malignant disease.

The bibliography is entirely concerned with American medical literature.

The Year Book of Cancer (1956–1957 Year Book Series). Compiled and edited by RANDOLPH LEE CLARK, jun., B.S., M.D., M.Sc. (Surg.), Director and Surgeon in Chief, the University of Texas M. D. Anderson Hospital and Tumor Institute; Professor of Surgery, the University of Texas Postgraduate School of Medicine; and RUSSELL W. CUMLEY, B.A., M.A., Ph.D., Houston, Texas, Director of Publications, the University of Texas M. D. Anderson Hospital and Tumor Institute. 7¾ × 5¼ in. Pp. 572, with 209 illustrations. 1957. Chicago: The Year Book Publishers (London: Interscience Publishers Ltd.). 57s.

THE *Year Book Series* is well known throughout the world and the books are popular and very informative. This is the first *Year Book of Cancer* and we would like to thank the compilers for a most useful reference book. Cancer appears to be on the increase in most parts of the world and this all-inclusive abstract volume should provide something that is lacking in the medical literature of to-day.

The illustrations and diagrams in this volume have been well chosen and add much to the usefulness of the text.

We look forward to seeing this annual volume next year.

A Text-book of X-ray Diagnosis. By British Authors, in three volumes. Edited by S. COCHRANE SHANKS, M.D., F.R.C.P., F.F.R., Director, X-ray Diagnostic Department, University College Hospital, London; and PETER KERLEY, C.V.O., C.B.E., M.D., F.R.C.P., F.F.R., D.M.R.E., Director, X-ray Department, Westminster Hospital; Radiologist, Royal Chest Hospital. Third edition. 10 × 7⅞ in. Pp. 521 + xiv, with 533 illustrations. 1957. London: H. K. Lewis & Co. Ltd. 90s.

THIS new third edition of this work has been very thoroughly revised and some new illustrations have been added.

The first volume, which contains five large parts, deals with the central nervous system, the teeth and jaws, the eye, the accessory nasal sinuses, and the ear and temporal bone.

It is an excellent volume with superb radiographs and illustrations.

This work is a great credit to British radiology and we look forward to seeing the other volumes which make up this magnificent work.

Aortography. Its Application in Urological and some other Complications. By W. BARR STIRLING, Ch.M., F.R.C.S. (Ed.), F.R.P.S.S.G., Senior Assistant, Urological Department, Glasgow Royal Infirmary; Consultant Urologist, Ministry of Pensions. 10 × 7 in. Pp. 292 + viii, with 155 illustrations. 1957. Edinburgh and London: E. & S. Livingstone Ltd. 50s.

THIS monograph is based on 500 aortograms carried out by the author in the Diagnostic Clinic of the Urological Department of the Glasgow Royal Infirmary.

There can be no doubt whatever that this book will convince the medical profession as a whole, and more especially urologists, of the real usefulness of this important diagnostic aid.

Although there are certain potential hazards associated with aortography, these are largely a matter of technique and are for the most part avoidable.

There are some 155 illustrations in this volume, the majority of which are aortograms, and every one has been beautifully reproduced. The advantage of aortography is that frequently definite pathological lesions of the kidney can be diagnosed with certainty—various congenital anomalies, renal cysts, neoplasms, hydronephrosis, calculus, tuberculosis of the kidney, and renal hypoplasia.

This book will take its place as one of the really worthwhile volumes which the clinician must have for reference purposes.

Mr. Barr Stirling has written a most useful book which is a real credit to British surgery.

Anesthesia and Otolaryngology. By DONALD PROCTOR, M.D., Assistant Professor of Laryngology and Otology, The Johns Hopkins University School of Medicine. 9 × 5¾ in. Pp. 268 + xvi, with 33 illustrations. 1957. Baltimore: The Williams and Wilkins Co. (London: Ballière, Tindall & Cox Ltd.). 56s.

THE author of this book is peculiarly well qualified to write it. After practising for many years as a laryngologist, he studied and practised anæsthesia for four years before returning, in 1955, to laryngology. In the practice of both specialities he was associated with some of the best and most progressive centres in the United States. His book is as good a blend of teaching and thought in the two disciplines as one could hope to find, and workers in both fields may read it with advantage, for it is full of good-humoured wisdom and practical common sense. The first of its four sections is devoted to general considerations, and includes chapters on preparation, premedication, and post-operative care. It includes some paragraphs on the post-operative recovery room and its manifold advantages, which all surgeons and anæsthetists in this country could read with profit. This is a matter on which thought in the United States is well ahead of our own. The second section deals in greater detail with otolaryngological procedures under general anæsthesia, and the third with those under local anæsthesia. The advantages, and the dangers, of local analgesia are excellently set forth. The final section is more discursive. It deals with respiratory resuscitation, high airway obstruction, problems of ventilation in the comatose patient, intrathoracic disease, and resuscitation of the newborn. Omissions are few. Congenital tracheo-œsophageal fistula, the treatment of which poses difficult problems for the anæsthetist and the surgeon, is barely mentioned; and nothing is said of the use of a cuirass respirator during bronchoscopy when muscle relaxants have been used.

The book is well produced, and it makes a valuable addition to the literature of its subjects. If anæsthetists fail to read it with the idea that it is written for surgeons, or surgeons with the belief that it is intended for anæsthetists, they will be missing a good thing.

Fundamentals of General Surgery. By JOHN ARMES GIUS, M.D., D.Sc. (Med.), F.A.C.S., Professor of Surgery, College of Medicine, State University of Iowa. 9 × 5½ in. Pp. 720, with 151 illustrations. 1957. Chicago: Year Book Publishers, Inc. (London: Interscience Publishers Inc.). 94s.

THIS book sets out to give the modern medical student just sufficient surgical knowledge to ensure that he can qualify at the end of his medical studies. The text is well written and the essential aspects of surgery have been well covered, but why is it necessary to list eighteen tests for liver function? Surely no student should be expected to know a fraction of these, and anyhow few of them are of any real value to the clinician.

The illustrations on the whole have been well chosen, but Fig. 119 on p. 560 demonstrates the lymphatics of the axilla draining into the supraclavicular triangle. This is contrary to British teaching, which postulates that the supraclavicular glands drain the anterior mediastinal lymphatics.

In spite of the few criticisms which have been mentioned this book will prove of value to the undergraduate student and should prove useful in ensuring a basic background of general surgery.

Operative Obstetrics. By R. GORDON DOUGLAS, Professor of Obstetrics and Gynecology, Cornell University Medical College; Obstetrician and Gynecologist in Chief, the New York Hospital; and WILLIAM B. STROMME, Attending Obstetrician and Gynecologist, Northwestern Hospital and Fairview Hospital, Minneapolis. 10 × 7 in. Pp. 735 + xiv, fully illustrated. 1957. New York: Appleton-Century-Crofts, Inc. $20.

THIS new work should prove to be popular because the authors have set forth in a very practical manner the important details of operative obstetrics. It is the detail which matters in operative surgery. The authors have also chosen the illustrations very carefully and the whole work is enriched by numerous photographs, line drawings, radiographs, and half-tone illustrations.

In no other work on this subject has there been such a wealth of illustrations.

There are answers to all the various queries which the young obstetrician is bound to conjure up in his mind when faced with difficulties. It is this very practical approach to the problem which makes this work so valuable.

A bibliography is given after each chapter and the various references have been carefully chosen.

An excellent work and a credit to American obstetrics.

Burns: Pathology and Therapeutic Applications. By SIMON SEVITT, M.D., M.Sc., M.A., F.R.C.P.I., D.P.H., Consultant Pathologist to the Birmingham Accident Hospital and M.R.C. Burns Research Unit. 8¾ × 5½ in. Pp. 364 + xii, with 85 illustrations. 1957. London: Butterworth & Co. (Publishers) Ltd. 37s. 6d.

MUCH has been written on the subject of burns during the last fifteen years, but this is the first book to be devoted to the important aspect of the pathology of burns. The author has had a wide experience of burns and has been able to interpret the significance of his pathological findings.

The book not only deals with thermal burns but includes electrical, chemical, and radiation burns.

In this age of rapid transport by road, sea, and air, burn casualties are all too common, and a book such as this will prove of immense value to surgeons, pathologists, and all practitioners who are concerned with the burned patient.

The illustrations in this monograph are excellent and well chosen. All medical students would do well to read this book.

Clinical Roentgenology of the Digestive Tract. By MAURICE FELDMAN, M.D., Assistant Professor of Gastroenterology, University of Maryland. Fourth edition. 10¼ × 7 in. Pp. 776 + xii, with 728 illustrations. 1957. Baltimore: The Williams & Wilkins Co. (London: Baillière, Tindall and Cox). 120s.

IT is nearly twenty years ago that the first edition of this work appeared and now the fourth edition has been published. During this period great advances in X-ray examination of the gastro-intestinal tract have taken place and to-day the diagnostic radiologist can give his medical and surgical colleagues much information regarding difficult abdominal conditions.

Dr. Feldman's clinical and radiographic experience enables him to write with authority and he methodically covers every phase of the gastro-intestinal tract from the œsophagus to the rectum.

The surgeon has come to rely considerably on the help he may obtain from his colleague in the X-ray department, and this book amply reveals that that help is forthcoming.

The large number of radiographs which illustrate this work are beautifully reproduced and if taken alone without the text would form an excellent atlas of gastro-intestinal conditions.

The references at the end of each chapter are full and comprehensive.

Clinical Gastroenterology. By EDDY D. PALMER, M.D., F.A.C.P., Lt. Col., Medical Corps, United States Army. 10 × 6¾ in. Pp. 630 + x, with 216 illustrations. London: Cassell & Co. 137s. 6d.

GASTRO-ENTEROLOGY is a large subject and one which has made great strides during the last few years. The modern X-ray diagnostic equipment has facilitated in no small way the great improvement in the diagnosis of gastro-intestinal lesions.

This large volume covers the whole gastro-intestinal tract in a very efficient manner and the standard of the many radiographs is very high.

As a work this book is comprehensive and covers not only the common but also the rare conditions of the alimentary tract. It should prove of value both to the busy general practitioner and to the advanced student in medicine.

Gastro-intestinal Obstruction. By MEYER O. CANTOR, M.D., M.S., F.A.C.S., Associate Attending Surgeon, Grace Hospital; Attending Surgeon, Sinai Hospital of Detroit; and ROLAND P. REYNOLDS, M.D., F.A.C.S., Chief of Surgery, Grace Hospital; and 3 other contributors. 10¼ × 7½ in. Pp. 566 + xii, with 415 illustrations. 1957. Baltimore: The Williams & Wilkins Co. (London: Baillière, Tindall & Cox Ltd.). 144s.

THIS is a very comprehensive work devoted to an important subject which has become even more important in the last decade.

There can be little doubt that the acute type of intestinal obstruction presents one of the most urgent diagnostic problems. But with gastric suction, modern anæsthesia with relaxants, and the antibiotics, and heparin, the surgeon to-day is in a better position to deal with these acute cases than ever before. The importance of fluid and electrolyte balance has also played a part in reducing the mortality rate of acute intestinal obstruction to about 10 per cent. This book, which is well illustrated, sets out in detail the various pre- and post-operative treatments and nursing care. The various forms of intestinal suction tubes are well described and the authors seem to favour the Cantor tube for intestinal decompression.

There is a comprehensive bibliography at the end of the book and a very practical index.

This large volume should interest the general practitioner, who after all is the first doctor to see these acute cases. The postgraduate student who is working for a higher surgical qualification will also find this book useful.

Operative Surgery. Edited by CHARLES ROB, M.C., M.Chir., F.R.C.S., Professor of Surgery, St. Mary's Hospital, London; and RODNEY SMITH, M.S., F.R.C.S., Surgeon, St. George's Hospital, London. 11 × 8½ in. Vol. 5, Part IX, Orthopædic Surgery. Pp. 1–366, fully illustrated. 1957. London: Butterworth & Co. (Publishers) Ltd. 110s. per volume.

THE fifth volume of this work is devoted to orthopædic surgery and covers every type of operative technique. The contributors are representative of those specializing in orthopædic surgery in the British Isles and all of them write on subjects that they have made their own. The whole volume is uniform with the preceding four volumes and the illustrations are uniformly good.

Perhaps the outstanding contribution comes from H. L. C. Wood, who writes on replacement arthroplasty of the hip and cup arthroplasty of the hip. The illustrations contained in this section are truly excellent.

This fifth volume is a great credit to British surgery and will take its place as a reference book for the postgraduate who wishes to take one of the higher examinations in surgery.

Peripheral Nerve Regeneration. Edited by BARNES WOODHALL, M.D., Professor of Neurosurgery, Duke University Medical School, Durham, North Carolina; and GILBERT W. BEEBE, Ph.D., Statistician, Follow-up Agency Division of Medical Sciences, National Research Council, Washington. 9¼ × 6 in. Pp. 671 + xxiv, with 60 plates. 1957. Washington: Veterans Administration.

THE Americans, like the British, have studied nerve injuries that occurred during the last war, but the plan of campaign was different in the two countries. The British organized five centres to which the injured combatants were sent at the earliest opportunity and where they were treated by special teams, who also carried out clinical and, in some places, experimental research at the same time. The British report was written entirely by those engaged in this work. The American study was not begun until 1946, one year after the end of the war, and it was, therefore, exactly as the title of this monograph says, a follow-up study. There was no connexion between those who had treated the patients and those who carried out this investigation. Furthermore, the work was done on a sample. The records of a little over 10,000 patients were available; the number examined was 2662, who presented 3656 nerve injuries. Distinguished teams of neurosurgeons and neurologists undertook this work in five centres—at Boston, Chicago, New York, Philadelphia, and San Francisco. They were chiefly concerned with the results of nerve suture and this was tackled with extraordinary thoroughness.

The conclusions reached were as follows, and it is of some interest to compare them with those arrived at by British workers: (1) The interval between injury and repair of the nerve had an influence on motor but not sensory recovery; the British found that both motor and sensory recovery were worse the longer the delay before

operation. (2) The larger the gap to be closed the worse the prognosis for motor but not for sensory recovery; in the British series the adverse effect applied to motor and sensory recovery alike. (3) Extensive mobilization of the nerve, required to close an extensive gap, was not found to have an adverse effect on motor recovery but, very surprisingly, it was followed by considerably poorer recovery of tactile sensibility in the lower limb though not in the upper. The British report does not deal with this point. (4) Bulb suture as a preliminary to definitive repair was not found to have an adverse effect. The British report says little on this because so few bulb sutures were performed. (5) The use of a stay suture, which in theory at any rate is harmful, may have had some adverse effect on motor recovery but had none on sensory recovery. (6) In the American armed forces it was fashionable to wrap tantalum foil round the suture line, and this was done in no less than a third of their cases. No definite conclusion was reached as to its value and in many cases the foil was removed later.

The report also contains valuable chapters on the electrical evidence of regeneration, on causalgia, and on functional recovery and occupational adjustment. There is a section on the correlation of the pathological findings—the histological state of the resected nerve-ends—and functional recovery. There ought to be a close correlation, but, as the British found, it was not clear cut.

This monograph is extremely difficult to read; not that it is badly written, but because the reader is overwhelmed by the statistical evidence. There are, indeed, no fewer than 286 tables, some of which are fairly complicated. This work cannot, therefore, be recommended to the surgeon who is anxious to learn something about the surgery of peripheral nerves. It is rather a work of reference for the man already expert in this field and for him it is invaluable.

Management of Complications in Eye Surgery. Avoidance of Pitfalls and Treatment of Difficult Situations in Ophthalmologic Operations. Edited by R. M. FASANELLA, M.D., Chairman of the Section of Ophthalmology, Yale University School of Medicine. 10 × 6½ in. Pp. 422 + xvi, fully illustrated. 1957. Philadelphia and London: W. B. Saunders Co. Ltd. 112s.

THIS large volume is devoted to the avoidance of pitfalls and treatment of difficult situations in ophthalmological operations. The various chapters are written by twenty contributors, each one dealing with his own speciality. The growth of ophthalmic knowledge and research in recent years has been rapid and fruitful.

The illustrations in this book depict actual operative technique and should prove of real value to those who wish to specialize in ophthalmic surgery.

It is a fact that the test of a good surgeon is his ability to deal with circumstances when things go wrong, and this book is full of ways and means of dealing with the unforeseen complications which may occur in ophthalmic operations.

Artificial Limbs. By LEON GILLIS, M.B.E., M.B., B.Ch., M.Ch.Orth., D.L.O., F.R.C.S., Consultant Orthopædic Surgeon to: Queen Mary's (Roehampton) Hospital, London; East Ham Memorial Hospital, London; St. John's Hospital, London. With a Foreword by Sir HARRY PLATT, LL.D., M.D., M.S., F.R.C.S., F.A.C.S. (Hon.). 9¾ × 7 in. Pp. 449 + xiv, fully illustrated. 1957. London: Pitman Medical Publishing Co. 300s.

IT is pleasing to see a sister text-book to Gillis's volume on *Amputations* which was hailed as the standard book on that subject.

The present work, devoted to the very important subject of artificial limbs, is just as masterly as the previous work. It explores by a single person the whole field of prosthetics, and presents a complete conspectus of all the surgical refinements and technical skill which have brought the fitting and making of artificial limbs to the present stage.

No one has had a better clinical background in orthopædic experience over the years than Gillis, and his vast actual observation at Roehampton is second to none. It is from this experience that the book has been written.

The book is divided into three parts and twenty-two interlocking sections, each of which can be studied independently.

Such a volume as this will be found invaluable not only to the surgeon, student, and technician, but also to all those numerous people who are concerned with rehabilitation of the maimed.

Every medical and public library should contain a copy of this comprehensive work.

BOOK NOTICES

[The Editorial Committee acknowledge with thanks the receipt of the following volumes. A selection will be made from these for review, precedence being given to new books and to those having the greatest interest to our readers.]

The Healing of Wounds. A Symposium of Recent Trends and Studies. Edited by MARTIN B. WILLIAMSON, Ph.D., Professor of Biochemistry, Stritch School of Medicine, Loyola University, Chicago. 8 × 5 in. Pp. 202 + xii, illustrated. 1957. New York: McGraw-Hill Publishing Company Ltd. 54s. 6d.

Anesthesia. A Manual for Students and Physicians. By STUART C. CULLEN, M.D., Chairman, Division of Anesthesiology, Department of Surgery, State University of Iowa Hospitals. Fifth edition. 8 × 5½ in. Pp. 295, with 37 illustrations. 1957. Chicago: The Year Book Publishers Inc. (London: Interscience Publishers Ltd.) 42s.

Traumatologia del Sistema Nervosa. By ANGELO CHIASSERINI. 9½ × 6½ in. Pp. 777, with 192 illustrations. 1957. Rome: Società Editrice Universo. L.9000.

Introduction to Dental Anatomy. By JAMES HENDERSON SCOTT, D.Sc., M.D., L.D.S., Reader in Anatomy for Dental Students, The Queen's University, Belfast, and NORMAN BARRINGTON BRAY SYMONS, M.Sc., B.D.S., Senior Lecturer in Dental Anatomy and Histology, and Curator of the Dental Museum, The University of St. Andrews at the Dental School, Dundee. 8¾ × 5½ in. Pp. 344+xii, with 219 illustrations. 1958. Edinburgh and London: E. & S. Livingstone Ltd. 42s.